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The Influence of Genetic Disorders on Parenting Stress and Family Environment

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The Influence of Genetic Disorders on Parenting Stress and Family Environment

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DEDICATION

This project is dedicated to my parents, Robert and Arlene Davis, and my brother and sister, Garth Davis and Toni Friedman. These four individuals have been my support, my inspiration, my motivation, and my source of all things wise and all things comical throughout my life. It is through them that I have found strength in times of weakness, humor in times of disappointment, and compassion in times of frustration. They are the foundation of my desire to help others. I also could not have completed this project with my sanity intact without the infinite encouragement of Christopher Scott, who has taught me more about motivation and perseverance than I could ever learn in 5 years of graduate school, and whose insight continues to instill hope in every corner of my world.

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The Influence of Genetic Disorders on
Parenting Stress and Family Environment

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18q- is a chromosomal deletion disorder caused by missing genetic material from the long arm of the 18th chromosome. The extensive impairments associated with 18q- may be a significant source of stress to parents. Research on families of handicapped children suggests that these families experience additional stress related to challenges such as increased caregiving demands, changes in social support systems, and financial burdens related to medical needs and decreased income. Changes in the family environment are also implicated in families coping with a disabled child. Some studies

reveal highly cohesive environments within these families, while others reveal decreased levels of expressiveness and cohesion and increases in conflict.

The present study compared variables of parenting stress and family environment in families of children with and without disabilities. Group 1 consisted of 24 primary caregivers of children with 18q-. Group 2 consisted of 32 primary caregivers of children with DS. Group 3 consisted of 32 primary caregivers of typically developing children.

A one-way, between groups multivariate analysis of variance (MANOVA) was conducted to investigate differences in parenting stress on three subscales of the Parenting Stress Index. A significant difference between groups was found. Post hoc pairwise comparisons indicated that the DS group reported statistically significantly more stress than the Control group on both the Isolation and Spouse subscales. The 18q- group was not found to be statistically significantly different from either the Control or DS group on any of the three PSI subscales.

A one-way, between groups multivariate analysis of variance (MANOVA) was also conducted to investigate differences in family environment on three subscales of the Family Environment Scale. A significant difference between groups was found. Post hoc pairwise comparisons indicated that the DS group showed statistically significantly less amounts of cohesion in the family environment than both the 18q- and Control groups. The 18q- group showed similar levels of cohesion to the Control group. There were no significant differences between groups on the other two FES subscales. Findings from the study provide important information about the role of family environment and

parenting stress in families of children with disabilities. Limitations of the study and implications for future research and practice are discussed.

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CHAPTER 1: INTRODUCTION

The 18q deletion is a chromosomal deletion disorder caused by missing genetic material from the long ("q") arm on the 18th chromosome. Research regarding this disorder has primarily focused on the physiological aspects of the disorder. Associated symptoms vary greatly in range and severity, but typical characteristics include mental retardation, delayed growth, hypotonia, microcephaly, and hearing impairment. Although the research is more limited, behavioral difficulties have also been documented, including hyperactivity, aggression, and autism.

The extensive impairments associated with this disorder, both physical and behavioral, may be a significant source of stress to parents of 18q- children. Research focused on stress related to parenting children with mental retardation, genetic disorders, and pervasive developmental disorders has revealed high amounts of anxiety regarding both the maladaptive behavior of these children, as well as the effects of such disorders on the family's environment and relationships (Beckman-Bell, 1981; Byrne & Cunningham, 1985; Crnic, Friedman, & Greenberg, 1983; Seligman & Darling, 1997). To date, there have been no studies that have evaluated family functioning in 18q-.

Research on families of handicapped children has suggested that these families experience additional stress related to specific challenges such as increased demands of caregiving, changes in social support systems, and financial burdens related to medical needs and decreased parent income because one parent may need to stay home to care for the child. In particular, the marital relationship may be affected by the significant changes that take place in a family coping with a child's disability, which in turn may

have a significant effect on the child's well-being (Floyd & Zmich, 1991; Howes & Markman, 1989). Additionally, parenting competence has been shown to be influenced by children's disabilities and to have an affect on stress levels in the family, with higher levels of stress being associated with decreased feelings of parental self-efficacy (Coleman & Karraker, 1997; Gross, Fogg, & Tucker, 1995; Jones & Prinz, 2005). Given the difficulties associated with parenting handicapped children, social isolation may also be a consequence of the diagnosis and result in increased parenting stress. Research shows that informal social support in particular is important in maternal feelings of competence and satisfaction (Haldy & Hanzlik, 1990; Kazak & Marvin, 1984; Van Hooste & Maes, 2003).

Changes in the family environment are also implicated in families coping with a disabled or handicapped child. In the relationship domain, research has been inconsistent, with some studies demonstrating highly supportive, cohesive environments within families coping with disabled children (Mahoney & O'Sullivan, 1992; Pueschel & Myers, 1994; Van Riper, Ryff, and Pridham, 1992). Other studies, however, have revealed decreased levels of expressiveness and cohesion, with increases in conflict within the family environment (Margalit & Heiman, 1986; Margalit & Raviv, 1983). Further research is needed in this area to determine the effects of a disabled child on the family environment and relationships between family members.

The present study examined and compared several variables of parenting stress and family environment in families of children with and without disabilities. Using data from 88 primary caregivers, the study compared such variables with families of children

with Down Syndrome, as well as with families of typically developing children. Group 1 consisted of 24 primary caregivers of children with 18q-. Group 2 consisted of 32 primary caregivers of children with DS. Group 3 consisted of 32 primary caregivers of typically developing children.

Given the high demands of caring for children with disabilities, this study hypothesized that for both the 18q- and the DS groups, the Competence, Isolation, and Spouse subscales of the Parenting Stress Index would be elevated compared to control subjects. A one-way, between groups multivariate analysis of variance (MANOVA) was conducted to investigate differences in parenting stress on three subscales of the Parenting Stress Index. A significant difference between groups was found. Post hoc pairwise comparisons indicated that the DS group reported statistically significantly more stress than the Control group on both the Isolation and Spouse subscales. The 18q- group was not found to be statistically significantly different from either the Control or DS group on any of the three PSI subscales.

Given the high level of involvement of many parents in families with disabilities this study also hypothesized that the levels of cohesion and expression on the Family Environment Scale would be elevated for the 18q- and DS groups compared to control subjects, while the levels of conflict were predicted to be lower in these two groups. A one-way, between groups multivariate analysis of variance (MANOVA) was conducted to investigate differences in family environment on three subscales of the Family Environment Scale. A significant difference between groups was found. Post hoc pairwise comparisons indicated that the DS group showed statistically significantly less

amounts of cohesion in the family environment than both the 18q- and Control groups. The 18q- group showed similar levels of cohesion to the Control group. There were no significant differences between groups on the other two FES subscales. Findings from the study provide important information about the role of family environment and parenting stress in families of children with disabilities. Limitations of the study and implications for future research and practice are discussed.

CHAPTER 2: LITERATURE REVIEW

This integrative analysis presents a review of chromosomal abnormalities, including a discussion of Down Syndrome and Chromosome 18 disorders. A discussion of types of parenting stress, family environment, and their implications in families with handicapped children will follow. A review of family assessment measures will be included, with focus on the two measures that were utilized in the current study. Finally, a rationale for the current study will be proposed.

Chromosomes

A chromosome can be described as a carrier for DNA, which contains the genetic material for making living organisms. Genes are segments of this DNA that are carried on each chromosome and determine specific human characteristics. Different kinds of organisms have different numbers of chromosomes. Humans have 23 pairs of chromosomes, 46 in all. Twenty-two of these pairs are numbered chromosomes called autosomes, and one pair are the sex chromosomes. Most cells in the human body contain the 23 pairs of autosomes, with each parent contributing one chromosome to each pair. Human reproductive cells, however, have 23 individual chromosomes rather than 23 pairs, so that when conception occurs, the fertilized egg will then have it's own 23 pairs of autosomes and one pair of sex chromosomes. For females, the sex chromosomes are two X chromosomes, and for males they are an X and a Y chromosome.

Each of the autosomes has been assigned a number from largest (1) to smallest (22). A karyotype, which is a display of the chromosomes of a single cell, is utilized to examine the chromosomes. Each chromosome has a black and white banding pattern and

a centromere, or waistband, in a specific location somewhere along is length. The centromere divides each chromosome into two unequal length arms. The shorter arm is called the "p" arm, and the longer arm is called the "q" arm. By looking at the location of the centromere and the black and white banding pattern on the karyotype, a person's chromosomes can be microscopically analyzed to determine if there are any aberrations. Abnormalities may include a reorganization of the material, a missing piece of material, or a duplication of material. When these types of aberrations occur, there may be severe consequences in an individual's physical and mental functioning.

Chromosomal Abnormalities

Chromosomal abnormalities are almost always associated with cognitive impairments, developmental delays, and various physical anomalies. The basic condition of mental retardation (MR) is a common problem independent of chromosomal abnormalities, with varying rates of prevalence depending on clinical definitions. When an MR diagnosis is based on intelligence alone, and based on the assumption of a normal distribution of intelligence in the population, it is expected that 3 percent of the population, or 6 million individuals in the U.S., are affected (Crandall, 1978; Murphy, Boyle, Schendel, Decouflé, & Yeargin-Allsopp, 1998). Because clinical definitions of MR generally require impairment of adaptive skills as well as intellectual difficulties, however, prevalence rates are generally estimated between 1 and 3 percent of the population (Crandall, 1978; Murphy et al., 1998). A review of the literature by Murphy et al. (1998) indicated that the prevalence of MR in children has ranged from as low as 1 per 1000 children to as high as 97 per 1000 children, with most of the variation in these

rates at higher IQ levels. According to the American Association on Mental Retardation (AAMR, 2002), the official definition of mental retardation is a disability characterized by significant limitations both in intellectual functioning and in adaptive behavior as expressed in conceptual, social, and practical adaptive skills that originates before the age of 18. In general, mental retardation is generally thought to be present if an individual has an IQ test score of approximately 70 or below. There are, however, numerous causes of MR and various levels of impairment (see Table 1.) According to the National Dissemination Center for Children with Disabilities (2004), about 87% of people with mental retardation will be slightly below average, between 50 and 75 on standardized intelligence tests, with the remaining 13% of people with mental retardation scoring below 50 on these tests.

Table 1. *Mental Retardation Classification Based on IQ Score*

Classification	**IQ Range**
Mild Mental Retardation	50-55 to approximately 70
Moderate Mental Retardation	35-40 to 50-55
Severe Mental Retardation	20-25 to 35-40
Profound Mental Retardation	< 20 or 25

Note. From Diagnostic and Statistical Manual of Mental Disorders – Fourth Edition, Text Revision (DSM-IV-TR), p. 49. Copyright 2000 by the American Psychiatric Association.

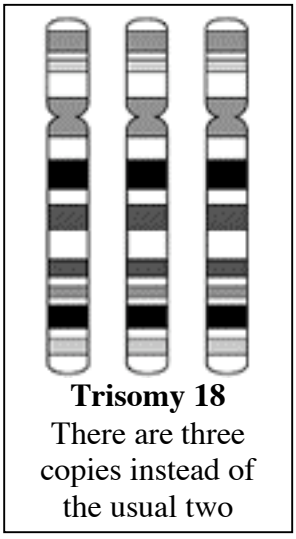
According to Crandall (1978), 50 percent of moderately and severely retarded individuals have a genetically related disorder. Chromosomal abnormalities account for 45 percent of these cases. More recent data from a review by Murphy, Boyle, Schendel, Decouflé, & Yeargin-Allsop (1998) revealed that genetic disorders account for

approximately 7-15% of all MR cases, and account for 30-40% of MR cases that are due to known causes. According to Murphy et al. (1998), chromosomal abnormalities account for up to 30% of severe MR cases and 4-8% of mild MR cases with identifiable causes. Plomin and Walker (2003) noted that more than 200 genetic disorders include mental retardation among their symptoms. According to the Chromosome 18 Registry and Research Society, most chromosomal abnormalities arise as de novo events, meaning that the error is a new one and occurred during the formation of the sperm or egg or very early in the embryonic development. Because they occur spontaneously, de novo events are not familial in nature, and parents of these children have a low probability of having another child with a chromosome abnormality.

In normal individuals, the correct number of human chromosomes is 46, with each cell having 22 pairs of autosomes and one pair of sex chromosomes. Chromosomal abnormalities can be numerical, where there is a loss or gain of one or more chromosomes. The most common case of a numerical chromosomal abnormality is called trisomy, which is associated with the addition of a single chromosome (see Figure 1). Chromosomal abnormalities can also be structural in nature, involving chromosome breakage (see Figure 2). One type of structural abnormality is a deletion, which results from a loss of part of the short (p) or long (q) arm of a chromosome. Deletions can vary by both size and location, thus clinical presentations of these disorders are variable. On chromosome 18, for instance, there are about 337 different genes. Deletions at any given point on the 18th chromosome can therefore result in a variety of symptoms. Even if two individuals are determined by karyotype to have the same deletion location, their

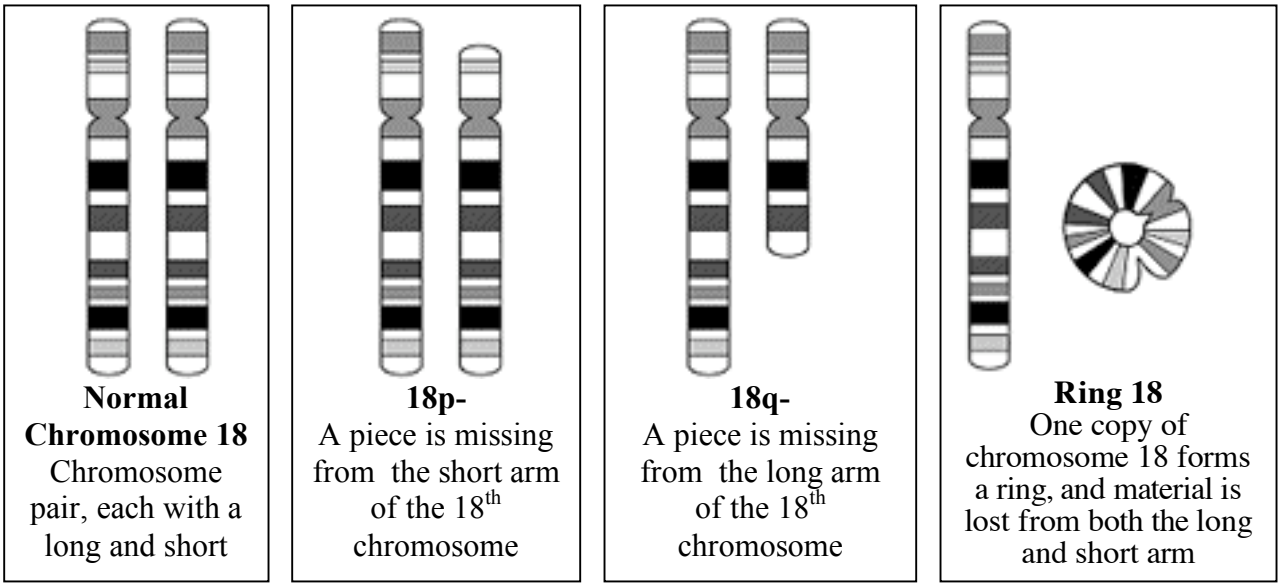
deletions could differ by as many as 50 genes. A ring chromosome is another structural abnormality that is formed when the broken ends of each arm of a chromosome fuse together. In this case, a variable amount of genetic material is lost on both arms. Deletion syndromes are most commonly found in chromosomes 9, 13, 21, 4, 5, and 18 (Crandall, 1978). These disorders are described as syndromes because they are generally associated with a pattern of recognizable physical anomalies, cognitive deficits, and behavioral dysfunction.

Figure 1. *Numerical Abnormality (trisomy) of Chromosome 18*



Note. From the Chromosome 18 Registry website (www.chromosome18.org).

Figure 2. *Structural Abnormalities of Chromosome 18*



Note. From the Chromosome 18 Registry Website (www.chromosome18.org).

Chromosome 18 Disorders

There are five major syndromes of chromosome 18. A wide variety of characteristics and severity exists across the disorders and even within each one. The five most frequent abnormalities of chromosome 18 include 18p- (deletion on the short arm, p), 18q- (deletion on the long arm, q), Ring 18 (one copy forms a ring), Tetrasomy 18p (presence of an extra chromosome made up of two copies of the short arm), and Trisomy 18 (three copies of the chromosome). The deletion syndromes of chromosome 18 (18q-, 18p-, and ring 18) occur in approximately 1 in every 46,000 births (the Chromosome 18 Registry and Research Society, 1991).

This study will focus on the functioning of families of individuals with 18q-. There is no evidence that 18q- disorder is caused by advanced maternal age or exposure to environmental agents. Additionally, there is no evidence that one ethnic group or

geographic area is more at risk. Individuals with 18q- typically have a normal lifespan, although it has been reported that approximately 10% of affected infants die in infancy (Pueschel & Thuline, 1983). The incidence of 18q- appears to be higher in females, with the female to male ratio at approximately 1.7/1.

Typically, the deletions in individuals with 18q- are terminal, meaning that the missing piece is at the end of the chromosome. Interstitial deletions, where the missing piece is in the middle of one of the arms, may occur but are generally less common. Because the size and location of chromosomal deletions vary, the clinical presentation of these syndromes is somewhat inconsistent; however, a pattern of recognizable physical anomalies, cognitive deficits, and behavioral dysfunction are often the diagnostic indicators of a syndrome. Individuals with 18q- generally exhibit poor growth, cognitive impairment, speech difficulties, microcephaly, CNS dysmyelination, hypotonia, and hearing impairment (Pueschel & Thuline, 1983; Chromosome 18 Registry and Research Society, 1991). These individuals may also exhibit behavioral difficulties, including hyperactivity, aggression, and autistic features (Mahr et al., 1996). Although these may be the most noted clinical features, a wide variety of symptoms are associated with 18q- (See Table 2).

Table 2. Major Clinical Features Found in Individuals with 18q Deletions

Characteristic	**% of Individuals**
Dysmyelination of the central nervous system	97
Speech failure	91
Hypotonia	79
Decreased or absent deep tendon reflexes	76
Foot deformities	74
Hearing loss	70
Mental retardation (IQ <70)	68
Gait abnormalities	68
Growth hormone deficiency	68
Proximally placed thumbs	65
Atretic/Stenotic ear canals	64
Tremor	62
Short stature	61
Microcephaly	53
Optic nerve hypoplasia	23
Autistic features	20
Nystagmus	14

Note. From the Chromosome 18 Clinical Research Center at the University of Texas Health Science Center at San Antonio, 2000.

Because the majority of individuals with 18q- have some degree of mental retardation (Chromosome 18 Registry and Research Society, 2000), adaptive behavior skill levels are generally significantly delayed and thus hinder the individuals' abilities to live independently. Cognitive delays vary in severity among individuals with 18q-, with reported IQ scores ranging from severely delayed (<40) to low average (88) on standardized intellectual tests such as the Wechsler Intelligence Scale for Children (Mahr et al., 1996; Wechsler, 1991). Dysmyelination of white matter has been reported throughout much of the brain in individuals with 18q-, which may be one cause of cognitive delays (Miller et al., 1990). A distinct neuropsychological profile of individuals with 18q- is uncertain due to limited testing involving children. The study by Mahr et al. (1996) indicates moderate to severe deficits in cognitive flexibility, executive functions, attention, novel problem solving, memory, language, visuomotor integration, and fine motor dexterity; however, in examining its implications for children, this study has some inherent limitations related to the age-appropriateness of some of the tests used.

Semrud-Clikeman, et al. (2005) found that in individuals with 18q- deletions, the degree of cognitive impairment is related to the amount of missing genetic material. Semrud-Clikeman et al. noted that those participants with the most missing genetic material obtained the lowest cognitive ability scores compared to participants with less missing genetic material, and they also tended to be the youngest. The Semrud-Clikeman et al. study has implications for the proposed study, especially in terms of the age-related impairment. If in fact younger individuals have larger deletions and thus greater

cognitive impairment, it may then be hypothesized that families of younger children with 18q- will experience added parenting stress and changes in the family environment.

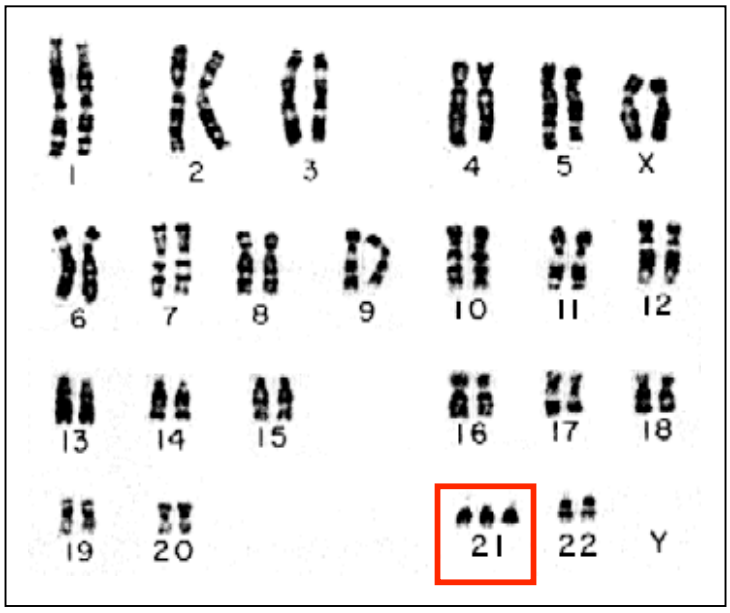
Down Syndrome

Down Syndrome (DS), the most common cause of developmental disability, is a chromosomal abnormality estimated to occur in 1 of every 700 live births (Selikowitz, 1997). Children with DS are usually developmentally delayed in terms of both physical and intellectual functioning. The cause of DS is unknown, although some have speculated that maternal age (above 35 years) may be a contributing factor. Other possible causes include hormonal abnormalities, genetic predisposition, or viral infection; however, there is no evidence that any of these conditions are directly responsible for the occurrence of DS (Pueschel, 1984). Many health concerns accompany the diagnosis of DS. Some of these include hearing deficits, congenital heart disease, intestinal abnormalities, eye problems, thyroid dysfunction, skeletal/muscular difficulties, and, later in life, Alzheimer's disease (Pueschel & Sustrová, 1997).

While DS is discussed in the literature as early as the 1500s, Dr. John Langdon Down was the first to identify the syndrome in 1866. DS is seen in all ethnic groups, and it is slightly more prevalent in boys than in girls. Physical features of DS are distinct and are important in making a diagnosis; however, it is essential to recognize that features vary across the population, and that some features may be present in individuals without a DS diagnosis. Selected features that may or may not be present include flattening of the back of the head, slanting of the eyelids, depressed nasal bridge, small mouth and ears, and decreased muscle tone (Selikowitz, 1997).

There are three types of DS. As noted above, Trisomy 21 is the most common form of DS, occurring in approximately 95% of cases (Selikowitz, 1997). People with Trisomy 21 have an entire extra 21st chromosome in every cell of the body (See Figure 3). Approximately 4% of DS cases are due to translocation, which is the presence of an extra piece of the chromosome rather than the whole chromosome. Mosacism accounts for approximately 1% of DS cases and involves the presence of a whole extra 21st chromosome in only a proportion of cells in the body. In other words, individuals with mosaic Down Syndrome have Trisomy 21 (and thus 47 chromosomes) in some of the cells in their body, while the rest of the cells in their body have the typical number (46) chromosomes.

Figure 3: *Karyotype for an individual with Trisomy 21 (Down Syndrome)*



Note. From the Riverbend Down Syndrome Parent Support Group website, http://www.altonweb.com/cs/downsyndrome/index.htm.

Children with DS vary in their rate of development, but in general they mature at a slower rate than the average child. Areas particularly affected in individuals with DS are speech and hearing, cognitive abilities, and motor skills. In addition, children with DS show difficulties in social development. Although their development in these areas may be retarded in comparison to the typically developing child, advances in early intervention programs have improved the potential for success in children with DS. Depending on environmental conditions and access to early intervention programs, children with DS may show cognitive abilities ranging from borderline to profound mental retardation (Hines & Bennett, 1996). In general, cognitive abilities continue to develop during infancy and early childhood, but they do so at a slower rate than a typically developing child.

Similar to the rate of development of cognitive abilities, children with DS show slower than average rates of motor development (Hines & Bennett, 1996). Infants with DS usually have hypotonia, meaning they show less muscle tone than typical, healthy infants and may be described as "floppy" (Hanson, 1988). Language development is also impaired in individuals with DS. Research indicates that language production in children with DS is particularly delayed as compared with their other cognitive abilities, including language comprehension (Miller, 1999). According to Hines and Bennett (1996), a variety of causes may contribute to the difficulties experienced in expressive speech. Some of these causes that commonly occur in children with DS may include craniofacial abnormalities, chronic respiratory infections, middle ear infections and impaired hearing, and deficits in vocal imitation skills, all of which result in indecipherable or distorted

speech. In particular, DS children show significant deficits in expressive language relative to their mental-age-matched peers.

A stereotype exists that children with DS are joyful, happy, carefree individuals. These characteristics may certainly be features of any human, however, and should not be projected onto each individual with this disorder. Although social skills are less affected than the delayed development often seen in other areas in individuals with DS, there are a number of distinctions in the social interactions of children with DS that demonstrate how they differ both from typically developing children and from the "happy and social" DS stereotype. Limitations to social interactions for these children include limited play repertoire, less initiative play behaviors, less use of eye contact for communicative purposes, and more repetitive acts (Hines & Bennett, 1996).

It is clear that both 18q- and Down Syndrome have significant effects on many aspects of functioning, but particularly on the cognitive and adaptive skills of individuals with these disorders. These significant effects have implications for families with children who have such disabilities. Research has indicated that families with handicapped children experience significant amounts of stress and changes in the family environment due to the unusual circumstances that raising a child with a disability entails.

Parenting Stress

According to family stress theory, the operation and functioning of families results from the complex interactions of external influences and the unique characteristics of individual family members (Mahoney & O'Sullivan, 1992). Proponents of family stress theory suggest that if problem-solving strategies are not utilized, the family will

continue in a state of disequilibrium (Minnes, 1988). In this state, roles are confused, needs are unmet, and goals are interrupted, and the management of resulting stress and crises depends on the nature of the circumstances as well as the nature of the individual family members.

Abidin (1992) noted that his first attempt to create an integrative model of parenting designated stress as the foundation of dysfunctional parenting behavior, where higher levels of parenting stress led to increases in dysfunctional parenting. His continued research revealed, however, that a "simple linear relationship did not exist between stress level and dysfunctional parenting" (Abidin, 1992, p. 408). Based on Lazarus and Folkman's transactional model of stress (1984), Abidin's more evolved model of parenting stress examined the influence of several sociological, environmental, behavioral, and developmental variables as they relate to a parent's self-expectations and beliefs about his or her "parenting role." In this model, parenting stress is a result of each parent's complex appraisals of his or her commitment to the parenting role (Abidin, 1992). Abidin's Parenting Stress Index (1995) allows researchers to examine these constructs from a self-report perspective.

Abidin's model of parenting stress is not specifically geared towards parents of children with disabilities, but the experiences of such families would likely correspond well with his underlying theory of self-appraisal as it relates to the parenting role. In other words, a parent's self-expectations and beliefs about his or her parenting role may become skewed when coping with difficulties in a child's development. Children with

disabilities have unusual characteristics that are likely to affect the psychological status of the family members and patterns of family functioning.

Research on families of handicapped children has suggested that these families experience additional stress related to certain characteristics of the child, including slower rate of development, less social responsiveness, more difficult temperament, more repetitive, stereotypic behavior patterns, and additional or unusual caregiving demands (Beckman-Bell, 1981; Beckman, 1983). According to Crnic, Friedrich, and Greenberg's 1983 review of the literature, families with disabled children must face significant challenges. Some challenges these families must cope with include increased demands of caregiving, changes in social support systems, and financial burdens related to medical needs and decreases in parent income. A study by Spangenberg and Theron (2001) revealed that almost one quarter of parents of children with Down Syndrome in their study were depressed, and that nearly half of the parents experienced above average anxiety levels. Walker, Van Slyke, and Newbrough (1991) found that parents of children with mental retardation obtained higher scores on scales that assess stress related to child caretaking demands as compared with control families of typically developing children.

Beckman and Pokorni (1988) noted that families of pre-term infants in particular experience a great amount of stress initially because of the child's fragile medical state and uncertain prognosis. They hypothesized that the initial stress may disappear as the child grows older and thus family stress would decrease over time. They found, in general, a significant decrease in the number of child problems reported at 12 months and another significant decrease at 24 months. Families of children with other disabilities

detected before or shortly after birth, such as genetic disorders, may show a similar pattern of stress. According to Seligman and Darling (1997), the first suspicion of disability and subsequent diagnosis are likely the most difficult times for new parents. They note that pregnancy and birth are stress-producing situations even when a baby is healthy, and that the additional fear and uncertainty that comes with a medical diagnosis exacerbates an already intense transition period.

Seligman and Darling (1997) also noted that parents of infants with disabilities may have more difficulty forming an attachment with their newborns than parents of infants without disabilities. They reported that when infants smile, make eye contact, and respond to parental attempts to feed and cuddle them, parents feel rewarded. Infants with disabilities are often unable to respond to their parents' efforts, however, leading to possible feelings of detachment or failure. According to Seligman and Darling, some characteristics of children with disabilities that may impede the formation of a secure parent-child attachment include feeding difficulties, medical fragility, inability to maintain eye contact, presence of medical equipment (such as feeding and oxygen tubes), excessive crying and fussiness, and a negative response to being handled.

Marital Relationship

According to literature regarding families of children with disabilities, the marital relationship may be at risk in these homes. Bristol, Gallagher, and Schopler (1988) reported that fathers of developmentally disabled children assumed less responsibility and were less involved in the care of the child than fathers of nondisabled children. They also reported that parents of disabled children reported significantly more marital difficulties

than their peers with nondisabled children. Notably, Bristol et al. reported that the functioning of the mothers of disabled children in terms of depression, marital adjustment, and parenting appeared to be related to their husbands' capacity to be supportive both instrumentally and expressively.

Howes and Markman (1989) reported that the ability of parents to handle differences in their relationships through appropriate conflict management and communication skills contributes to their child's well-being. Their longitudinal study revealed that predictive associations were found between the quality of the mother's relationship with her husband before marriage and later child security of attachment and sociability. It has been noted throughout the literature that marital distress and dysfunction may add stress to the family and thus disrupt the parent-child bond (Emery, 1982; Floyd & Zmich, 1991). Marital difficulties can have negative long term repercussions for child development (Emery, 1982; Howes & Markman, 1989). A 1982 review of the literature by Emery revealed that several studies have found relationships between unhappy, conflict-ridden marriages and child behavior problems (Hetherington, Cox, & Cox, 1976; Porter & O'Leary, 1980; Wallerstein & Kelly, 1976; as cited in Emery, 1982). Emery noted that throughout the literature on children in families with discord and divorce, child behavior problems have included delinquency, conduct problems, anxiety-related problems, depression, aggression, and demandingnesss.

Floyd, Gilliom, and Costigan (1998) noted that the parenting alliance, the component of marital relationships that pertains specifically to parenting together, is one important factor in successful parenting experiences. It has been reported that marital

difficulties may undermine this parenting alliance and thus may disrupt effective parenting (Belsky, 1984; Jouriles & Farris, 1992). Floyd et al. (1998) found that in families of children with mental retardation, couples with positive marriages reported greater confidence in their own parenting competence, showed improvements in feelings of parenting competence over time, and tended to reduce negative interactions with their children over time as compared to couple with poorer marital functioning. Other studies of mothers of children with mental retardation have demonstrated that spousal support is predictive of lower stress scores (McKinney & Peterson, 1987). A study by Floyd and Zmich (1991) revealed that relatively severe child behavior problems in children with mental retardation were associated with reports of lower marital satisfaction by both fathers and mothers. A review article by Morgan, Robinson, and Aldridge (2002) examined parenting stress in relation to externalizing child behavior. According to this review, there is a general consensus in the literature that parenting stress involves a mismatch between perceived resources and the actual demands of the parenting roles.

Crnic, Friedrich, and Greenberg (1983) noted in their review of the research that studies of marital satisfaction do not reveal a uniform and consistent pattern, and that marital response is likely dependent upon factors aside from the presence of a child with mental retardation, such as severity of the handicap, age and sex of the child, and the quality of the marital relationship prior to the birth of the child. The authors of the review did note, however, that previous studies generally show that parents of children with mental retardation have clinically significant profiles compared with norm groups on the Minnesota Mutli-phasic Personality Inventory (MMPI) (Erickson, 1968, 1969;

Miller & Keirn, 1978). Byrne and Cunningham (1985) reported that the high levels of stress mothers of mentally handicapped children experience appear to be more related to subjective factors such as their feelings of restriction and social isolation rather than to directly measurable features such as divorce rates. They concluded that the lack of consistent findings in this area suggests that the measurement of basic demographic and structural differences between families provides insufficient information to distinguish between those families who are subject to high levels of stress and those who are not.

Another review of the literature by Benson and Gross (1989) indicated that the majority of studies regarding marital relationships in families with handicapped children are inconclusive. According to Benson and Gross (1989), several studies have reported a significantly higher divorce rate in these families as compared to families with typically developing children (Breslau & Davis, 1986; Leyendecker, 1982; Stevenson, Graham, & Dorner, 1978; Tew, Lawrence, Payne, & Rawnsley, 1977; as cited in Benson & Gross, 1989). In general, however, Benson & Gross noted that the majority of studies do not find such differences. Results of these studies may be inconclusive, but it does appear that the presence of a handicapped child has a significant impact upon the marriage: very few studies in the Benson and Gross review reported that the child had not affected the marital dyad at all.

Benson and Gross (1989) did note that many studies report that the stresses and burdens associated with parenting a handicapped child have decreased marital satisfaction and deteriorated the marital relationship, including poor communication, financial problems, and sexual difficulties. Still, they also reported that in some studies,

parents of handicapped children experience greater marital satisfaction and cohesiveness as a result of the child's presence. Benson and Gross's review concluded that the vast majority of studies focus on more severe and well known disorders, such as cystic fibrosis, spina bifida, and Down Syndrome, and that those of lesser severity or that are less prevalent are rarely studied in the literature. Benson and Gross suggested that those parents of children with mild or marginal handicaps may have more difficulty accepting their children's limitations. It is also possible that less information and support exists for families coping with less severe handicaps, and thus these families do not have the same resources that families with more severely handicapped children have. The lack of research on these families of children with mild or marginal disabilities, however, makes it difficult to come to such conclusions.

Parenting Competence

Parenting competence, or parental self-efficacy, involves the degree to which parents see themselves as effective in their parenting role. According to Bandura's definition of the construct, self-efficacy refers to the belief in one's ability to successfully perform a particular behavior (Bandura, 1977). The Bandurian framework maintains that self-efficacy does not represent a global, fixed personality trait, but that rather it changes in response to different tasks and situations. Other theories of self-efficacy depart from Bandura's traditional model, viewing the construct as a stable personality trait or a general sense of efficacy across many behavioral domains (Coleman & Karraker, 1997).

In their review of the literature, Coleman and Karraker (1997) noted that according to the Bandurian approach, in order for parents to feel efficacious, they must possess knowledge of child care responses, have confidence in their own abilities to carry out such tasks, and believe that their children will respond contingently and that others in their social and family system will be supportive of their efforts. In general parenting self-efficacy has not been studied in great depth, but Coleman & Karraker noted that despite this deficiency, the existing studies demonstrate the importance of this construct for understanding personal satisfaction, adjustment to parenting, and the quality of the environment that parents are able to provide for their children.

In their 2005 review, Jones and Prinz cited several instances of a negative correlation between parenting self-efficacy and stress (Gross, Fogg, & Tucker, 1995; Erdwins, Buffardi, Casper, & O'Brien, 2001; Scheel & Rieckmann, 1998). These studies generally concluded that decreased parenting stress is related to increased feelings of parenting self-efficacy. Still other studies in the Jones and Prinz review revealed a positive relationship between parents' satisfaction and their self-efficacy.

According to the Coleman and Karraker (1997) review, low maternal self-efficacy has been correlated with maternal depression, maternal defensive and controlling behaviors, actual behavior problems in children, high levels of maternally reported stress, and a passive coping style in the parenting role. They reported that based on the parenting self-efficacy literature, "it is possible to assert that high parenting self-efficacy is strongly related to maternal ability to foster a healthy, happy, and nurturant childrearing environment" (Coleman & Karraker, 1997, p. 62).

A study by Hastings and Brown (2002) revealed that parents of children with autism have high levels of potential mental health problems. The authors noted that fathers in this study with high self-efficacy were less anxious than were those with low self-efficacy when the child had a high level of behavior problems. Gowen, Johnson-Martin, Goldman, and Appelbaum (1989) conducted a study with mothers of handicapped and nonhandicapped infants. Despite the additional caregiving demands that the mothers of handicapped infants must deal with, the study found that the mean levels of depression and parenting competence were not significantly different from the mothers of nonhandicapped infants. The authors did find, however, that a higher percentage of the mothers of handicapped infants scored at or above the cutoff point for risk for clinical depression than did mothers of nonhandicapped infants, supporting the general consensus in the literature that higher levels of maternal depression are related to the greater difficulty involved in caring for a handicapped child. There are a lack of studies examining the relationship between parental self-efficacy in parents of children with intellectual disabilities and/or developmental delays. The studies on this topic that do exist indicate that self-efficacy in parents of children with intellectual disabilities may be predictive of parental stress (Friedrich, Wilturner, & Cohen, 1985; Frey, Greenberg, and Fewell, 1989).

Social Isolation

The role of social relationships in coping is somewhat uncertain in the literature, and little research exists that looks formally at the nature of social support networks in families with handicapped children. Caplan and Killilea (1976) noted that social support

helps to accomplish adaptation in three ways: providing emotional mastery, offering guidance regarding problems and methods of coping with them, and providing feedback on behavior that fosters improved performance. Social networks may be formal in nature, such as professional health care programs, or informal, such as family and friend involvement. In their 1984 review of the literature, Kazak and Marvin noted that informal social networks appear to be more critical to family adjustment and adaptation.

According to Wikler (1981), individuals with mental retardation have historically been stereotyped in the public. Wikler noted that people generally feel uncomfortable with mentally handicapped individuals and strive to avoid interacting with them. Kazak and Marvin (1984) pointed out that neighbors may be perceived as being reserved towards the family, and when offered, help tends to be directed towards nonhandicapped siblings. In his chapter on interventions with parents of individuals with mental retardation, Tymchuk (1983) reported that the predominant historical view of those with mental retardation has been one of "worthlessness," with the mentally retarded individual being seen as less than a person based on intellectual deficiency and physical deformity (p. 370). Tymchuk further described the view of the economic implications of individuals with MR who are unable to care for themselves. Woolfson (2004) also noted the view of disability as a "personal tragedy" that has dominated in our society, and that this view leads us to believe it would be better if disabled people had not been born. Woolfson goes on to discuss the societal implications of the different approaches to disabled individuals, including disability as a medical problem, disability as a tragedy, and disability as a barrier to independence.

The view of disability as a tragedy for both the parents and the child is most notable in relation to social isolation. Woolfson (2004) describes feelings of pity that friends and family may demonstrate towards families of children with disabilities and noted that effective parents must choose to disregard such feelings and maintain a positive view to help the child develop into a worthwhile member of the community. Efforts have been made to amend society's negative views and give individuals with mental retardation the rights that other individuals receive. With the advent of early intervention programs, the general demise of institutionalization, and the least restrictive environment clause of the Individuals with Disabilities Education Act (IDEA, 1990), handicapped individuals have more opportunities than ever to experience the same stimulating and challenging environments as their typically-developing peers. Negative stereotypes of individuals with disabilities do remain, however, and it is these stereotypes and feelings of pity that may add to the social isolation and lack of social support that families of handicapped individuals may endure.

Few studies examine the amount of social support that families of children with disabilities receive compared to families with typically developing children, but a study by Williams, Elder, and Griggs (1987) indicated that of families of 60 children with developmental disabilities, 27% reported that they lacked a support system within the family, while 33% reported that they lacked a support system outside the family such as relatives and friends. Gayton (1975) and McAndrew (1976) also revealed diminished social support. McAndrew reported that a third of the parents in her study claimed that caring for a disabled child put restrictions on the number of outings they were able to go

on together. One third of parents in the McAndrew study reported that they believed their disabled child brought about adverse changes in their relationships with friends and extended family, particularly in the early months of the child's life.

A study by Friedrich (1979) found that social support was not significantly related to coping; however, other research generally reveals a complex relationship between the two. Gayton (1975) indicated that families with a handicapped child tend to experience social isolation. Other studies have revealed that parents report feeling that their relationships with friends and family were adversely affected by the birth of a handicapped child (McAndrew, 1976). In a study by Kazak and Marvin (1984), parents of children with spina bifida had significantly smaller friendship networks than parents of non-handicapped children, although these networks were considered to be more closely-knit than those in the control group. According to the authors, however, although the closely-knit networks may foster cohesiveness and support, they also generate stress in relation to diminished access to outside resources and viewpoints. Waisbren (1980) found similar results in her study, which revealed that the presence of higher degrees of family involvement is associated with increased levels of marital discord and increased individual stress reactions. In general, Kazak and Marvin reported that the larger the social network, the greater the likelihood of successful coping and adaptation.

According to McKinney and Peterson (1987), research indicates that mothers have often identified the emotional support they receive from other mothers as one of the most important aspects of group meetings and participation in other early intervention type programs. In their study, McKinney and Peterson did not find a significant

relationship between social support and stress level; however, over half of their intervention subjects mentioned peer support and the interaction with other mothers as the most helpful aspect of their early intervention programs. A study by Haldy and Hanzlik (1990) demonstrated that adequate social support had a positive influence on maternal feelings of competence. In their review of the literature regarding family factors of children with Down Syndrome, Van Hooste and Maes (2003) concluded that social support positively affects family functioning, quality of parenting style, parental attitudes, and parents' perceptions of their child.

Kazak and Marvin (1984) reported that the development and maintenance of friendships are based on sharing common interests and activities, which is difficult for families with handicapped children based on the special demands that these children place on the family. Minnes (1988) found that social support from extended family, friends, and neighbors was negatively correlated with the stress of mothers of children with mental retardation. Similarly, Beckman (1991) found that for both mothers and fathers of children with disabilities, increased informal support was significantly associated with decreased stress. A study by Beckman and Pokorni (1988) that examined stress in families of preterm infants also found that stress was significantly negatively correlated with informal support. Van Hooste and Maes (2003) also noted the importance of informal social networks and the emotional support and information about parenting that they may offer. The general conclusions among these studies are that informal support, such as from families and friends, is important to the well-being of families coping with children with disabilities. It also appears that more formal supports,

such as health care based programs, are not correlated with family stress (Beckman, 1991; Beckman & Pokorni, 1988; Minnes, 1988; Van Hooste & Beas, 2003).

Family Environment

Moos and Moos' (1976) model of family environment assesses family climate in terms of three domains: personal growth, system maintenance, and relationships. Moos and Moos developed the Family Environment Scale (FES), a parent self-report, true-false measure that assesses these three underlying domains (Billings & Moos, 1982; Moos & Moos, 1976). Various research has indicated that higher degrees of personal growth and supportive relationships within the family are linked with increased adjustment and fewer physical and emotional symptoms (Margalit & Heiman, 1984; Margalit & Raviv, 1983; Margalit, Raviv, & Ankonina, 1992).

Billings & Moos (1982) utilized the FES to classify families based on family typologies. They found that cohesion and expressiveness are key characteristics in families classified as "support-oriented." The authors noted that these families emphasized interpersonal relationships rather than specific areas of personal growth and goal attainment, and also that these families experienced the fewest stressful events. Billings & Moos reported that husbands in these support-oriented families experienced high levels of social interaction, while wives reported experiencing considerable support in their work settings. Conflict-oriented families, on the other hand, were high on control and conflict scales, but low on the interpersonal scales of expressiveness and cohesion. These families had the highest incidence of stressful events.

Several studies have concluded that although parenting stress may be increased by the presence of a chronically ill child, the basic dimensions of family functioning are not necessarily disrupted (Cadman, Rosenbaum, Boyle, & Offord, 1991; Kazak, 1987; Walker, Van Slyke, & Newbrough, 1992). Walker et al. (1992) noted that family conflict was no greater in families of children with chronic illness than in families of typically developing children. In their study of families of children with Down Syndrome, Van Riper, Ryff, and Pridham (1992) found that families with a child with Down Syndrome are more comparable to than different from families of nondisabled children. Dyson (1991) noted that although stress in families of handicapped children is elevated, this stress does not appear to be predictive of family dysfunction. Dyson concluded that families generally appear to respond to the care of a handicapped child with resilience and adaptive functioning despite the presence of family stress.

Mahoney and O'Sullivan (1992) conducted a study utilizing the Family Environment Scale (Moos, 1974) to examine the family environments of children with disabilities. Their results indicated that families of children with disabilities participated less in recreational activities and had a stronger moral-religious orientation than the general population. Mahoney and O'Sullivan also found, however, that the more severe the child's handicap, the more likely families were to have FES scores suggestive of distressed family functioning. Given that individuals with 18q- generally have a high degree of impairment, this is an important finding. Because of the severity of their handicaps, it is possible that the family environment of children with 18q- might be more distressed than the family environment of children with other, less severe disabilities.

Mahoney and O'Sullivan concluded that in general, the interpersonal relationships as measured by cohesion, conflict, and expressiveness appeared slightly more favorable for families of children with disabilities than for the normative sample of families.

Pueschel and Myers (1994) also utilized the FES to study family environments and revealed similar results to Mahoney and O'Sullivan's. In their study of the family environments of children with Down Syndrome, Pueschel and Myers found that high scores on the FES were observed in the cohesion, expressiveness, achievement, moral/religious emphasis, organization, and control categories. They concluded that the above-average scores on the cohesion and expressiveness scales indicate a high degree of commitment and support family members provide for one another as well as the extent to which members are encouraged to express their feelings.

Not all studies have found higher amounts of expressiveness and cohesion in families of handicapped children. A study by Margalit and Raviv (1983) found that mothers of children with mental retardation viewed their families as less encouraging of open expression of emotions. Another study by Margalit and Heiman (1986) found a decrease in the expression of emotion and the cohesiveness of the family system as a whole related to the presence of a learning disabled child. Blacher, Nihira, and Meyers (1987) administered the FES to families of mildly, moderately, and severely mentally retarded children. They found that families of the children with severe mental retardation had the lowest scores of the groups on all of the subscales. All three groups had low average levels of conflict compared to the FES norms. Additionally, all three groups had average levels of cohesion and expressiveness compared to the FES norms.

The research examining family environment generally focuses on family climate as a whole. The few studies that examine individual subscales of the FES, such as expressiveness, cohesion, and conflict, provide inconsistent findings. Although some studies have found no significant differences across FES domains (Dyson, 1991; Van Riper, Ryff, and Pridham, 1992), other studies revealed increased expression and cohesion (Mahoney & O'Sullivan, 1992; Pueschel & Myers, 1994), while still other studies revealed impaired family functioning (Blacher, Nihira, and Meyers, 1987; Margalit and Heiman, 1986; Margalit and Raviv, 1983). More research is needed in this area in order to establish a pattern of family climate among families coping with handicapped children.

Measurement and Assessment in the Family System

Various family assessment measures have been developed over time, based on both theory and intervention. One method often utilized to assess family environment and functioning is observation, during which an administrator observes a family's interactions and utilizes a coding system to track relevant behaviors and interactions. Although this method may provide a more detailed, third person examination of a family, the observation method is time consuming, vulnerable to rater and inter-rater reliability problems, and requires extensive training of administrators to rate the families.

Self-report measures are another method utilized to assess family environment and functioning. Self-report measures are questionnaire formats that individual family members complete. These questionnaires reveal a family member's subjective

experience in a family system. The ease of administration and completion of these measures makes them a frequent method of family assessment.

Self-Report Measures

Parenting Stress Index. The Parenting Stress Index (PSI; Abidin, 1995) is a self-report measure that provides an estimate of areas of stress in parent-child relationships. The PSI measures three main subscales, the Parenting Domain, Child Domain, and Life Stress. The measure is completed by the primary caregiver about a specific child and typically takes about 30 minutes to complete. The PSI is comprised of 120 items that are used to form composite scores on Child, Parent, Total Stress, and Life Stress domains. For the purpose of validity, the measure also includes a Defensive Responding scale to assess the degree to which the respondents present themselves favorably or minimize the problems or levels of stress in the parent-child dyads. The items are measured on a 5 point Likert scale ranging from 1 (strongly agree) to 5 (strongly disagree). Percentile scores are used to interpret responses: scores from the 15^{th} to 80^{th} percentiles are considered within the normal range. For this study, the scores on the Competence, Isolation, and Spouse scales of the parent domain of the PSI will be evaluated to determine the level of parenting stress.

The PSI was normed on 2633 parent-child dyads. The PSI has been empirically validated across a variety of ethnicities. Reliability estimates for the PSI range from .55 to .80 on the parent domain in the standardization sample. Test-retest reliability for the parent domain of the PSI ranges from .69 to .91. Additionally, the PSI appears to be valid and correlates well with other measures of parenting stress.

Family Environment Scale. The Family Environment Scale (FES) is a widely used questionnaire that has been applied in research with a diverse array of groups, including families of alcoholics, families of children with cystic fibrosis, and families of different nationalities (Rousey, Wild, & Blacher, 2002). Although it was not developed specifically for use with families of children with mental retardation, the FES has been utilized to examine the environments of families with developmentally delayed children (Boyce, Behl, Mortensen, & Akers, 1991; Blacher, Shapiro, Lopez, Diaz, & Fusco, 1997; Rousey and Rogers-Dulan, 2000, as cited in Rousey et al., 2002).

The FES is a parent self-report, true-false measure that focuses on the interpersonal relationships among family members, the directions of personal growth emphasized in the family, and the family's organizational and system-maintenance characteristics (Billings & Moos, 1982). The measure contains nine subscales with 10 items each. Together, these nine subscales assess three main dimensions of the family environment: relationship (cohesion, expressiveness, conflict); personal growth (achievement orientation, intellectual-cultural orientation, active recreational orientation, moral religious emphasis); and system maintenance (organization, control). The concurrent and predictive validity of the FES have been empirically validated (Moos & Moos, 1986). In this study, three FES subscales from the relationship domain, Cohesion, Expressiveness, and Conflict, will be analyzed separately. Subscale scores were standardized to a mean of 50 and standard deviation of 10 using the data from the authors' preliminary normative sample of 285 families (Moos & Moos, 1976).

Summary and Rationale

The 18q- disorder, a chromosomal deletion disorder caused by missing genetic material from the long ("q") arm on the 18th chromosome, has significant effects on the individuals with the disorder. Although research regarding this disorder has primarily focused on the physiological aspects of the disorder, there are several associated symptoms that are additionally stressful for both the individual with 18q- and his/her caretakers. Additional symptoms of 18q- vary greatly in range and severity but include mental retardation, delayed growth, hypotonia, microcephaly, and hearing impairment. Although the research is more limited, behavioral difficulties have also been documented, including hyperactivity, aggression, and autism.

The previous literature review has demonstrated that the extensive impairments associated with 18q-, both physical and behavioral, may be a significant source of stress to parents of 18q- children. Research that has focused on stress related to parenting children with mental retardation, genetic disorders, and pervasive developmental disorders has demonstrated high amounts of anxiety regarding both the maladaptive behavior of these children, as well as the effects of such disorders on the family's environment and relationships (Beckman-Bell, 1981; Byrne & Cunningham, 1985; Crnic, Friedman, & Greenberg, 1983; Seligman & Darling, 1997). To date, there have been no studies that have evaluated the family variables in the 18q- disorder.

Stress related to specific challenges such as increased demands of caregiving, decreases in social support systems, and financial burdens related to medical needs and changes in parent income has been found in the research on families of handicapped

children. Particular areas of stress that may be affected in families coping with a child's disability include the conflict in the marital relationship, decreased parenting competence, and increased social isolation. The corresponding subscales of the Parenting Stress Index can directly measure each of these areas.

Changes in the family environment are also implicated in families coping with a disabled or handicapped child. Research has been inconsistent, with some studies demonstrating highly supportive, cohesive environments and other studies revealing decreased levels of expressiveness and cohesion, with increases in conflict within the family environment. Further research is needed in this area to determine the effects of a disabled child on the family environment and relationships between family members.

It is clear that parenting stress and family environment have been studied extensively in the literature, although no studies have been found that examine these constructs within families of children with 18q-. Research on families of children with disabilities demonstrates that these families exhibit high levels of parenting stress, martial discord, social isolation, and decreased parent feelings of self-efficacy. The evidence regarding family environment is conflicted, and it is unclear what changes in expressiveness, cohesiveness, and conflict occur in families of disabled children. The significant impairments that children with 18q- have in intellectual ability and adaptive behavior, along with the medical complications that often accompany this diagnosis, suggest that these children place higher levels of stress on their parents and likely affect the family environment and relationships. It is important to note that more severe handicaps have been associated with more distressed family functioning (Mahoney &

O'Sullivan, 1992), thus the extensive impairments associated with 18q- suggest that these families may experience additional stress and difficulties within the family as compared to control groups.

The current study enhances previous research on parenting stress and family environment and examines these variables in families of children with 18q-. This study contrasts parenting stress and family environments of families with 18q- children with a clinical control group of families with Down Syndrome children and a control group of families with typically developing children. Given the demands of caring for a child with 18q-, this study hypothesized that for all clinical subjects, the selected subscales of the Parenting Stress Index would be elevated compared to both groups of control subjects. Given the high amount of involvement of many parents in families with 18q- children, this study hypothesized that the levels cohesion and expression on the Family Environment Scale would be elevated compared to control subjects, while the levels of conflict are predicted to be lower. The 18q- disorder is far less common than Down Syndrome; thus, it is likely there are not as many local support groups and resources available for parents and families coping with 18q-. Additionally, children with 18q- have a high incidence of medical difficulties that may add stress to the family environment. This study hypothesized that the levels of cohesion and expression on the FES would be elevated compared to the clinical control subjects with Down Syndrome, while the levels of conflict were predicted to be lower. Additionally, the study hypothesized that all three of the PSI scales would be elevated for the 18q- group compared to the clinical controls.

CHAPTER 3: METHOD

Chapter 3 is divided into four major sections: *Participants, Procedures, Instrumentation,* and *Data Analyses*. The *Participants* section includes demographic information and the process of recruitment for group selection. The second section, *Procedures,* describes the procedures used for data collection. The *Instrumentation* section includes descriptions of the measures, including their reliability and validity information. The fourth section, *Data Analyses,* describes the statistical analyses used, as well as the hypotheses and rationales.

Participants

Demographics

This study included 88 participants who are the primary caregivers of children between the ages of 1 month and 6 years old. Participants were recruited to be a part of three groups: a clinical group and two control groups. The clinical group included 24 primary caregivers of children who have been diagnosed with a deletion on the long arm of the 18th chromosome. This group included 20 mothers, 3 fathers, and 1 grandmother. One control group included 32 primary caregivers of children who have been diagnosed with Down Syndrome. This group included 29 mothers, 2 fathers, and 1 grandmother. The second control group included primary caregivers of typically developing children, as determined by a brief questioning of developmental history. The typically developing control participants were excluded if the developmental history revealed notable neurological, medical, or psychiatric difficulties. This control group included 32

mothers. The groups approximated each other in ethnicity and an attempt was made to match the groups on gender and age. Information was also gathered from primary caregivers regarding marital status. Demographic information for the sample is presented by group in Table 5 in the Preliminary Analyses section of Chapter 4 and includes caregiver's status (mother, father, grandmother), child's age and gender, caregiver ethnicity, and marital status. English was the primary language for all participants.

Recruitment

The clinical group of participants came from the Chromosome 18 Clinical Research Center at the University of Texas Health Science Center in San Antonio, an ongoing study investigating the clinical and educational effects of chromosome 18. These participants are primary caregivers who have children that were previously identified to have a deletion on the long arm of the 18th chromosome (18q-). The control group including primary caregivers of typically developing children was recruited from community samples. The second control group including primary caregivers of children with Down Syndrome was recruited with the help of contacts at Down Syndrome of Louisville, the Down Syndrome Association of Central Texas, and the Down Syndrome Association of San Antonio. The researcher's contacts sent an informative email about the study to members of their associations. Participants volunteered to join the study by emailing the researcher or their contact at the associations with their address and child's age. Letters, consent forms, and packets containing directions, the questionnaires, and a pre-addressed, pre-stamped envelope were mailed to potential participants.

This study complied with the ethical issues and standards of research set forth by the American Psychological Association and the University of Texas at Austin. Approval for the use of human subjects for the clinical participants in the Chromosome 18 study was obtained from the Institutional Review Boards of the University of Texas Health Science Center in San Antonio. Research materials were also submitted to the Departmental Review Committee within the Department of Educational Psychology and the Institutional Review Boards of the University of Texas for approval of the use of the control participants.

Because this study recruited the clinical participants from the larger ongoing research project as well as local agencies, we utilized a convenience sampling method, rather than a random sample of the general population.

Procedures

Data Collection

Families of children with 18q- that participate in the larger Chromosome 18 study are provided with transportation arrangements and an itinerary detailing their procedure schedules as a part of the larger study. Participants generally stay for 4 days at accommodations located near the research facilities, during which time they complete a standard research protocol that includes genetic analysis, Magnetic Resonance Imaging (MRI), behavioral audiology exam, and neurodevelopmental testing.

The neurodevelopmental assessment for each participant in the Chromosome 18 study typically takes 2-2 ½ hours, including parent interview, cognitive testing, and screeners for adaptive and emotional/behavioral functioning. The assessment battery also

includes measures for the primary caregiver to complete that examine family environment and parenting stress. Letters inviting individuals to participate in the control aspect of this research project were e-mailed to the primary caregivers of children in the appropriate age group. Participants in the two control groups did not receive a neurodevelopmental assessment. The primary caregiver measures, along with a letter explaining the measures, a consent form, a contact number for the investigator, and a pre-addressed stamped envelope were sent out to those volunteers who met the criteria.

All data entry and analysis was completed using assigned coded identification numbers to prevent the use of identifying confidential information. All assessment files for the clinical group, including testing protocols and questionnaires, were secured in a locked file cabinet in the office of a principal investigator of the Chromosome 18 study, Dr. Margaret Semrud-Clikeman. All of the testing protocols and completed questionnaires for the control subjects were secured in a locked file cabinet in the student investigator's home office.

Administration and Scoring

The primary caregivers in the clinical group completed the self-report measures while their children completed the neurodevelopmental assessment with a trained school psychology doctoral student under the supervision of a licensed psychologist. Families who have participated in the Chromosome 18 study previously but did not complete these measures and primary caregivers in the control groups received the measures in the mail with a letter detailing the instructions for the measures, as well as the investigator's contact number in case they had questions. Upon completing the measures, the primary

caregivers used the pre-addressed, stamped envelope they received to return the completed measures to the investigator.

All primary caregiver measures for this study were scored by the student investigator using the scoring and norms outlined in the manuals for each of these measures. The Parenting Stress Index (PSI; Abidin, 1995) and the Family Environment Scale (FES; Moos, 2002) were not scored if more than 2 items on each subscale of interest are not answered in order to protect measure reliability. Additionally, if the Defensive Responding scale of the PSI was in the clinically significant range, these participants were excluded from the study. The measures and their subscales are described in more detail in the Instrumentation section of this document.

Instrumentation

The following dependent measures were collected from the primary caregivers: the Parenting Stress Index (PSI; Abidin, 1995) and the Family Environment Scale – Real Form (FES; Moos, 2002).

Parenting Stress Index

The Parenting Stress Index (PSI; Abidin, 1995) is a self-report measure that provides an estimate of areas of stress in parent-child relationships. The measure is completed by the primary caregiver about a specific child and typically takes about 30 minutes to complete. The PSI is comprised of 120 items which are used to form composite scores on Parent, Total Stress, and Life Stress domains. For the purpose of validity, the measure also included a Defensive Responding scale to assess the degree to which the respondents present themselves favorably or minimize the problems or levels

of stress in the parent-child dyads. The items are measured on a 5 point Likert scale ranging from 1 (strongly agree) to 5 (strongly disagree). Percentile scores are used to interpret responses: scores from the 15th to 80th percentiles are considered within the normal range. For this study, the scores on the Competence, Isolation, and Spouse scales of the PSI were evaluated to determine the level of parenting stress. Percentile scores were converted into Standard Scores with a mean of 100 and a standard deviation of 15 using a psychometric conversion table.

The PSI was normed on 2, 633 parent-child dyads. The PSI has been empirically validated across a variety of ethnicities. Reliability estimates for the PSI range from .55 to .80 on the parent domain in the standardization sample. Test-retest reliability for the parent domain of the PSI ranges from .69 to .91. Additionally, the PSI appears to be valid and correlates well with numerous other measures of parenting stress (Abidin, 1995). One study examined the Child Behavior Scale (CBS; Adams, 1982) in hearing parents of hearing-impaired children and found that the PSI Total Stress score was significantly correlated with the CBS results (Adams & Tidwell, 1989). Another study by Holden, Willis, and Foltz (1989) examined the relationship between the Child Abuse Potential Inventory (CAP; Milner, 1986), and found significant correlations between the Abuse Potential score on the CAP and several of the PSI subscale scores. Significant correlations between the PSI and several other measures of parenting stress have been documented in the literature, including the Inventory of Parent Experiences (IPE), the Maternal Social Support Index (MSSI), the Parenting Sense of Competence (PSOC), the Beck Depression Inventory (BDI), and the Family Resources Scale (FRS).

Family Environment Scale – Real Form

The Real form of the Family Environment Scale (FES-R, Moos, 2002) is a self-report measure that assesses a family member's perception of his own family environment. The Real form of the FES is completed by primary caregivers about their actual perceptions of their families' environments, and it typically takes about 30 minutes to complete. The FES is comprised of 90 true/false items which are used to form composite scores on 10 subscales that assess three underlying domains: Relationships, Personal Growth, and System Maintenance (Moos & Moos, 2002). T-scores are used to interpret responses: scores from 40 to 60 are considered within the normal range. For this study, the scores on the Relationship domain, including the Expressiveness, Cohesion, and Conflict subscales, were evaluated to examine the family environment.

The FES appears to be a valid measure of family environment. The cohesion subscale of the FES correlates with the Social Support Appraisals scale and the Social Support Questionnaire (Sarason, Shearin, Pierce, & Sarason, 1987; Vaux, Phillips, Holly, Thomson, Williams, & Stewart, 1986). Dickerson and Coyne (1987) noted that the cohesion and control subscales on the FES have been correlated with other self-report measures of family cohesion and control, including the Family Assessment Device (FAD) and the Family Adaptability and Cohesion Evaluation Scales (FACES II). Spiegel and Wissler (1983) conducted a study in which professionally trained staff members rated families of psychiatric patients based on information obtained during a home visit. The authors found that the staff members' ratings were correlated with family members' reports of cohesion, expressiveness, conflict, and religious emphasis. Internal

consistency reliability estimates for the Form R subscales of the FES range from .61 to .78. Test-retest reliability estimates suggest that the scale is stable across time intervals.

Data Analyses

Research Question 1

What are the levels of parenting stress in families of children with 18q- and Down Syndrome?

Hypothesis 1a. Primary caregivers of children with 18q- will show above average levels of stress related to competence, isolation, and spouse relationships. The families of children with 18q- will show higher levels of stress compared to those with families with Down Syndrome or typically developing children.

Hypothesis 1b. Primary caregivers of children with Down Syndrome will show above average levels of stress related to competence, isolation, and spouse relationships compared to the PSI norm group and the typically developing control group; however, these levels will be lower than the 18q- levels.

Rationale. It is expected that primary caregivers of children with 18q- will exhibit above average levels of parenting stress related to competence, isolation, and spouse relationships. Children with 18q- experience a multitude of difficulties, including medical complications, behavior problems, delayed cognitive abilities, and below average adaptive skills. It has been shown that stress in the family is elevated when children have more severe developmental and neurological problems (Beckman, 1991; Crnic, Friedrich, & Greenberg, 1983; Dyson, 1996). Because children with Down Syndrome also often exhibit medical complications, impaired cognitive abilities, and other developmental

delays, it is expected that parenting stress in these families will be similar to the families of children with 18q-. The DS group is expected to experience less stress than the 18q- group based on the prevalence of DS and the availability of resources available to DS families.

Data Analysis. First, a one-way analysis of variance (ANOVA) was conducted to determine if child ages were statistically significantly different between the groups. Because a statistically significant difference was found between the groups for age, a multiple analysis of covariance (MANCOVA) was conducted for the PSI and examined to determine if there were statistically significant differences between the groups while controlling for the effects age as a covariate. Because there were no significant differences between the groups when age was controlled for, a multiple analysis of variance (MANOVA) was used to determine if there was a statistically significant difference between the 18q- clinical group and the DS and typically developing control groups on the 3 dependent variables, the parenting competence, social isolation, and spouse relationship subscales of the PSI. These scores were compared between groups using the Wilks' Lambda criterion. A Tukey HSD post hoc analysis was also done on the data. Chapter 4 offers the detailed statistical analysis of the data, as well as graphs that give a visual representation of the results.

Research Question 2

What are the types of family relationships in families of children with 18q- and Down Syndrome?

Hypothesis 2a. Primary caregivers of children with 18q- will show above average levels of cohesion and expressiveness in their family environments. The families of children with 18q- will show higher levels of cohesion and expressiveness compared to those with families with Down Syndrome or typically developing children.

Hypothesis 2b. Primary caregivers of children with 18q- will show below average levels of conflict in their family environments. The families of children with 18q- will show lower levels of conflict compared to those with families with Down Syndrome or typically developing children.

Hypothesis 2c. Primary caregivers of children with DS will show above average levels of cohesion and expressiveness in their family environments compared to the FES norm group and the typically developing control group; however, these levels will be lower than the 18q- levels of cohesion and expressiveness.

Hypothesis 2d. Primary caregivers of children with DS will show below average levels of conflict in their family environments compared to the FES norm group and the typically developing control group; however, this level will be higher than the 18q- levels of conflict.

Rationale. It is expected that both 18q- and DS families will exhibit above average levels of cohesion and expressiveness, and below average levels of conflict. Children with 18q- and DS experience a multitude of difficulties, including medical complications, behavior problems, delayed cognitive abilities, and below average adaptive skills. Because children with DS exhibit some similar impairments as children with 18q-, it is expected that family environments will be similar to the families of

children with 18q-. Although evidence is mixed, some studies indicated that families with handicapped children show above average levels of cohesion and expressiveness, and below average levels of conflict (Mahoney & O'Sullivan, 1992; Pueschel & Myers, 1994). These studies indicate that families with handicapped children may experience a greater need to become more cohesive and supportive in the face of these difficulties. It is possible that because DS is more common and has become a well-known diagnosis with many resources available, DS is less likely to cause the intense supportive response that families coping with rarer, more severe disabilities experience. Based on this hypothesis, the DS group is expected to experience less cohesion and expressiveness and more conflict than the 18q- group.

Data Analysis. First, a one-way analysis of variance (ANOVA) was conducted to determine if child ages were statistically significantly different between the groups. Because a statistically significant difference was found between the groups for age, a multiple analysis of covariance (MANCOVA) was conducted for the FES and examined to determine if there were statistically significant differences between the groups while controlling for the effects age as a covariate. Because there were no significant differences between the groups when age was controlled for, a multiple analysis of variance (MANOVA) was used to determine if there was a statistically significant difference between the 18q- clinical group and the DS and typically developing control groups on the 3 dependent variables, the cohesion, expressiveness, and conflict subscales of the FES. These scores were compared between groups using the Pillai's Trace criterion. A Tukey HSD post hoc analysis was also done on the data. Chapter 4 offers

the detailed statistical analysis of the data, as well as graphs that give a visual representation of the results.

CHAPTER 4: RESULTS

The present study examined several variables of parenting stress and family environment in families of children with 18q-, Down Syndrome (DS), and typically developing children between the ages of 1 month and 6 years. Additionally, the present research provided a step toward a more in depth understanding of the types of parenting stress and family environment that occur in families of children with and without genetic disorders. This study investigated primary caregivers' ratings on two measures of parenting stress and family environment across 18q- and DS and across age in order to understand more about these constructs within families coping with genetic disorders. This section details the findings of the analyses presented in the previous chapter. Descriptive statistics are presented first, followed by preliminary analyses. The next section includes the results for each hypothesis. The final section summarizes the results.

Descriptive Statistics

Descriptive statistics are presented by group in Table 3 and include means and standard deviations for each subscale of each measure. Each subscale was statistically significantly correlated with the others, as shown by Table 4. Statistics are presented using standard scores with a mean of 100 and a standard deviation of 15 for the Competence, Isolation, and Spouse subscales of the Parenting Stress Index (PSI). Statistics are also presented utilizing T-scores with a mean of 50 and a standard deviation of 10 for the Cohesion, Expressiveness, and Conflict subscales of the Family Environment Scale (FES).

Table 3. *Means and Standard Deviations of Measures/Subscales by Group (N = 88)*

Measure/Subtest	**Control** (n = 32)		**18q-** (n = 24)		**DS** (n = 32)	
	M	SD	M	SD	M	SD
FES Cohesion [a]	61.06	8.09	61.25	9.42	52.50	15.16
FES Expressiveness [b]	60.63	10.48	56.83	11.76	53.84	13.96
FES Conflict [c]	43.50	9.07	43.92	12.55	49.75	14.27
PSI Competence [d]	94.59	13.58	92.46	19.18	100.41	13.82
PSI Isolation [e]	95.00	16.18	102.08	13.15	105.22	17.96
PSI Spouse [f]	104.59	11.26	109.71	12.1	112.50	11.69

[a] Family Environment Scale Cohesion subscale

[b] Family Environment Scale Expressiveness subscale

[c] Family Environment Scale Conflict subscale

[d] Parenting Stress Index Competence subscale

[e] Parenting Stress Index Isolation subscale

[f] Parenting Stress Index Spouse subscale

Table 4. Correlation Matrix for Subscales Using Pearson's Correlation

		FES Coh [a]	FES Exp [b]	FES Con [c]	PSI Com [d]	PSI Iso [e]	PSI Spo [f]
FES Coh [a]	Pearson Corr	1	.559 **	-.490**	-.435**	-.440**	-.485**
	Sig. (2-tailed)	.	.000	.000	.000	.000	.000
	N	88	88	88	88	88	88
FES Exp [b]	Pearson Corr		1	-.388**	-.331**	-.348**	-.337**
	Sig. (2-tailed)		.	.000	.002	.001	.001
	N		88	88	88	88	88
FES Con [c]	Pearson Corr			1	.314**	.289**	.427**
	Sig. (2-tailed)			.	.003	.006	.000
	N			88	88	88	88
PSI Com [d]	Pearson Corr				1	.305**	.367**
	Sig. (2-tailed)				.	.004	.000
	N				88	88	88
PSI Iso [e]	Pearson Corr					1	.381**
	Sig. (2-tailed)					.	.000
	N					88	88
PSI Spo [f]	Pearson Corr						1
	Sig. (2-tailed)						.
	N						88

** Correlation is significant at the 0.01 level (2-tailed).

[a] Family Environment Scale Cohesion subscale

[b] Family Environment Scale Expressiveness subscale

[c] Family Environment Scale Conflict subscale

[d] Parenting Stress Index Competence subscale

[e] Parenting Stress Index Isolation subscale

[f] Parenting Stress Index Spouse subscale

Preliminary Analyses

A test of normality was conducted on the data and revealed statistically significant skewness for each of the FES subscales . According to Stevens (2002), when sample size is equal, MANOVA is robust for violations of normality. Because the 18q- group had only 24 subjects, it was necessary to take a random sample of each of the other two groups in order to create equal groups of 24 subjects each. When the data were analyzed with equal group numbers, a statistically significant difference that was similar to the *F* value in the original data analysis was revealed, indicating that the MANOVA was in fact robust for the violation of normality. There was no statistically significant skewness evident in the normality test of the PSI subscales. Demographic information for the sample is presented by group in Table 5 and includes caregiver's status (mother, father, grandmother), child's age and gender, caregiver ethnicity, and marital status.

Table 5. Sample Demographic Data by Group (N = 88)

Demographic	Control (*n* = 32)	18q- (*n* = 24)	Down Syndrome (*n* = 32)
			n (%)
Caregiver Status			
Mother	32 (100.0)	20 (83.3)	29 (90.6)
Father	0 (0.0)	3 (12.5)	2 (6.3)
Grandmother	0 (0.0)	1 (4.2)	1 (3.1)
Caregiver Ethnicity			
Caucasian	27 (84.4)	23 (95.8)	25 (78.1)
African American	1 (3.1)	0 (0.0)	0 (0.0)
Latino	1 (3.1)	1 (4.2)	6 (18.8)
Asian American	0 (0.0)	0 (0.0)	0 (0.0)
Other	3 (9.4)	0 (0.0)	1 (3.1)
Caregiver Marital Status			
Married	31 (96.9)	20 (83.3)	27 (84.4)
Divorced	0 (0.0)	3 (12.5)	1 (3.1)
Separated	1 (3.1)	0 (0.0)	1 (3.1)
Single	0 (0.0)	1 (4.2)	3 (9.4)
Child Gender			
Male	18 (56.3)	13 (54.2)	9 (28.1)
Female	14 (43.8)	11 (45.8)	23 (71.9)
Child Age			
<12 months	14 (43.8)	0 (0.0)	4 (12.5)
12-23 months	3 (9.4)	2 (8.3)	4 (12.5)
24-35 months	6 (18.8)	3 (12.5)	5 (15.6)
36-47 months	4 (12.5)	1 (4.2)	4 (12.5)
48-59 months	0 (0.0)	7 (29.2)	8 (25.0)
60-71 months	3 (9.4)	6 (25.0)	4 (12.5)
71-82 months	2 (6.3)	5 (20.8)	3 (9.4)

A one-way analysis of variance (ANOVA) was conducted to determine if the age of the child was statistically significantly different between the groups. A statistically significant difference for age was found between groups, $F(2, 85) = 14.46$, $p \leq .01$, with the control subjects being statistically significantly younger than both the 18q- and the DS groups, and the 18q- group being significantly older than both the Control group and the DS group. Table 6 provides means and standard deviations for age by group. Because a statistically significant difference was found between the groups for age, a co-variate was added to the multiple analysis of variance (MANCOVA) for each measure (FES and PSI) to determine if there were statistically significant differences between the groups while controlling for the effects age as a covariate. The results of each MANCOVA revealed similar results to the MANOVA analyses, indicating that even when age was controlled for, the levels of statistical significance did not change. The tests of hypotheses will therefore include the results for the MANOVA analyses. It should be noted that because MANOVA uses repeated tests, the analyses have an inflated Type I error rate.

Table 6. Age Means and Standard Deviations by Group (N = 88)

	18q- (***n* = 24**)	**Down Syndrome** (***n* = 32**)	**Control** (***n* = 32**)
M (age in months)	56.33	42.63	26.00
SD	17.81	21.77	22.65

Results of Tests of Hypotheses

Hypothesis 1a and 1b

Hypothesis 1a predicted that parents of children with 18q- would show above average levels of stress related to competence, isolation, and spouse relationships compared to the norm group on the PSI, and that the families of children with 18q- would show higher levels of stress compared to the norm group compared to families with Down Syndrome or typically developing children. Hypothesis 1b predicted that parents of children with Down Syndrome would show above average levels of stress related to competence, isolation, and spouse relationship compared to the PSI norm group and the typically developing control group; however, these levels were expected to be lower than the 18q- levels. Three dependent variables were used: the Competence, Isolation, and Spouse subscales of the PSI. The independent measure was group (Control, DS, or 18q-). Preliminary assumption testing was conducted. The variables were not found to be statistically significantly skewed, and thus the assumption of normality was not violated. Box's test for homogeneity of variance-covariance matrices was examined and was not found to be significant. According to Mertler and Vannatta (2002), Wilks' Lambda may be used as the test statistic if Box's test is not found to be significant.

Compared to the PSI norms, the group means for each of the three PSI subscales were within the average range. Using the Wilks' Lambda criterion, the results of the one-way between-groups MANOVA showed a statistically significant multivariate effect for groups on the combined dependent variables, multivariate $F(6,166) = 2.217$, $p = .044$. Analysis of Variance (ANOVA) was conducted on each dependent variable as a follow-up test to MANOVA. In examining the univariate effects of the dependent variables, there were statistically significant differences between the groups on the PSI Isolation

subscale, $F(2, 85) = 3.35$, $p = .04$. Statistically significant differences between the groups on the PSI Spouse subscale were also found, $F(2, 85) = 3.77$, $p = .027$. Differences between groups on the PSI Competence subscale were not significant, $F(2, 85) = 2.08$, $p = .131$.

The Tukey HSD post hoc analysis revealed that there were statistically significant differences between the DS group and the Control group on two of the three PSI subscales. According to the post hoc analyses, the DS group reported statistically significantly more stress than the Control group on the PSI Isolation subscale ($p = .035$). The post hoc analyses also revealed that the DS group reported statistically significantly more stress on the PSI Spouse subscale than the Control group ($p = .022$). Although statistical significance was found for these two subscales, they were not in the direction as predicted by hypotheses 1a or 1b. The 18q- group was not found to be statistically significantly different from either the Control group or the DS group on any of the three PSI subscales. Figure 4 demonstrates the expected results of the mean group differences on the three PSI subscales. Figure 5 demonstrates the observed results of mean group differences on the three PSI subscales.

Figure 4. *Expected PSI Subscale Means*

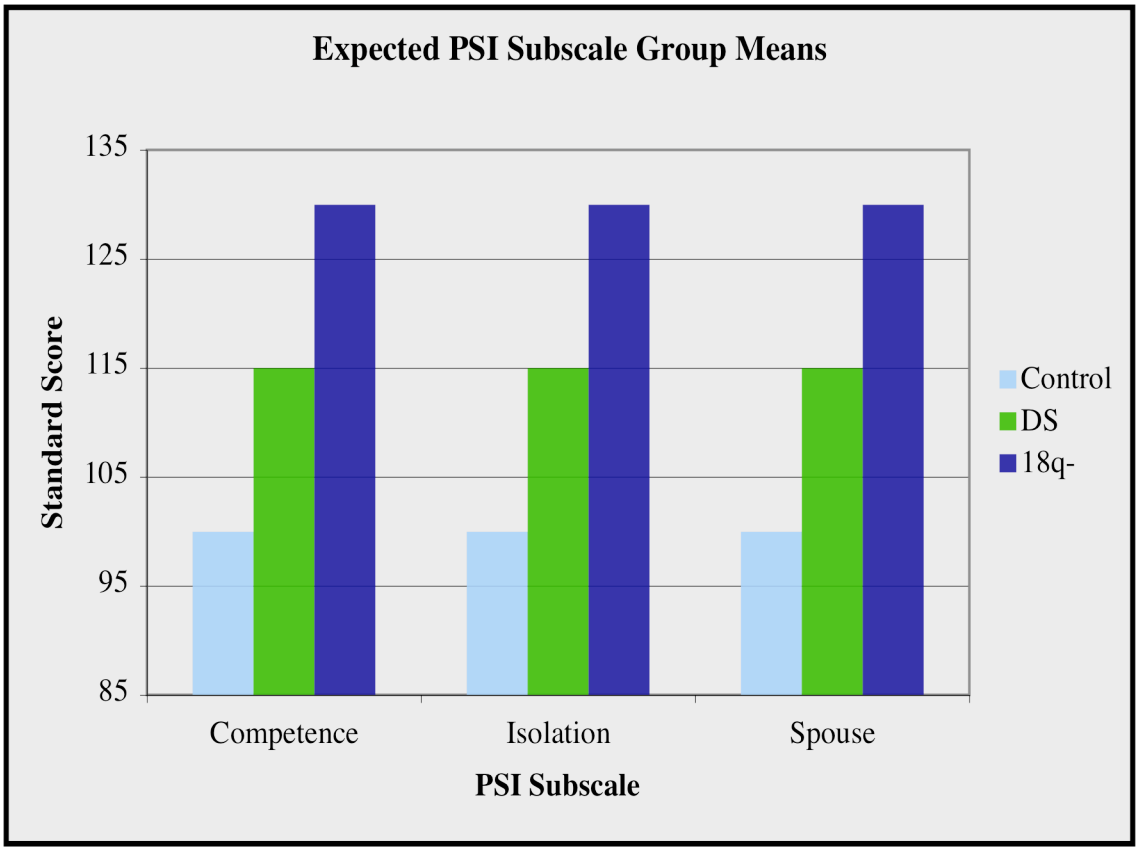
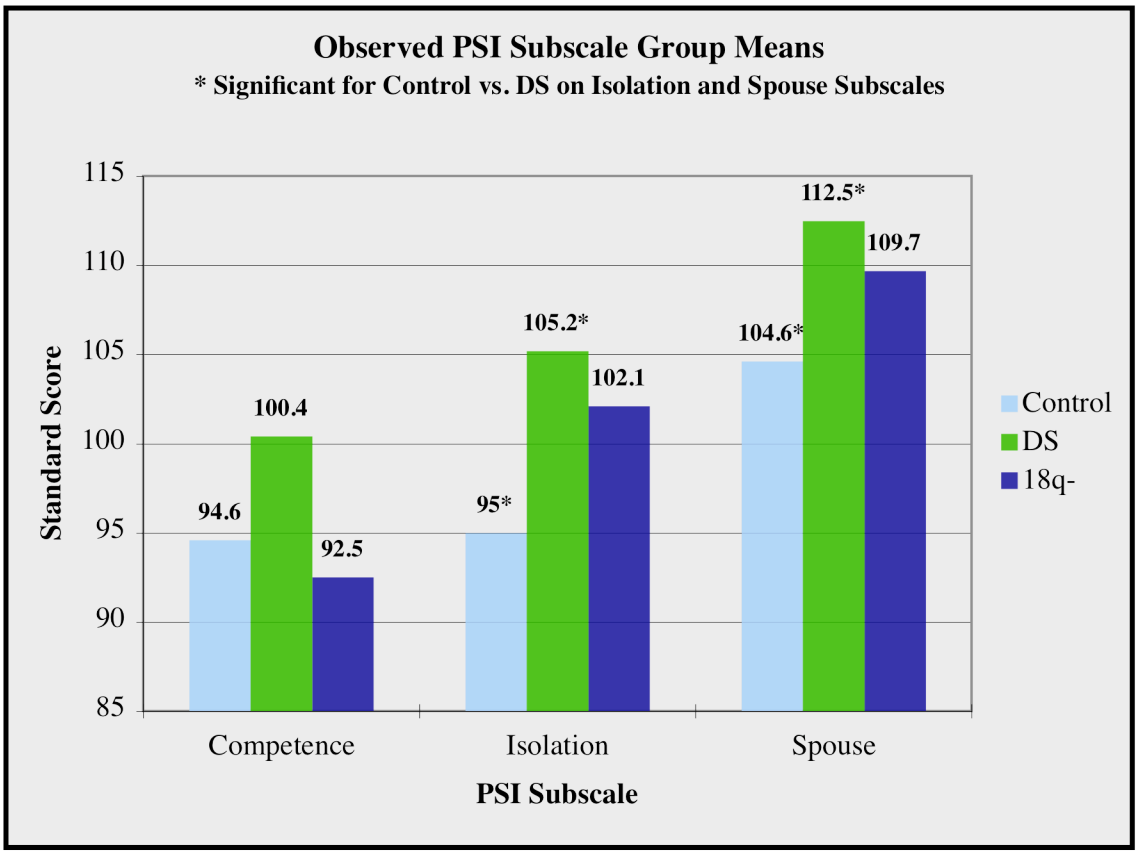


Figure 5. *Observed PSI Subscale Means*



Hypotheses 2a, 2b, 2c, and 2d

Hypothesis 2a predicted that parents of children with 18q- would show above average levels of cohesion and expressiveness in their family environments. The families of children with 18q- were also expected to show higher levels of cohesion and expressiveness compared to those with families of children with Down Syndrome or typically developing children. Hypothesis 2b predicted that parents of children with 18q- would show below average levels of conflict in their family environments and that the families of children with 18q- would show lower levels of conflict compared to those with families with Down Syndrome or typically developing children. Hypothesis 2c

predicted that parents of children with DS would show above average levels of cohesion and expressiveness in their family environments compared to the FES norm group and the typically developing control group; however, these levels will be lower than the 18q- levels of cohesion and expressiveness. Hypothesis 2d predicted that parents of children with DS would show below average levels of conflict in their family environments compared to the FES norm group and the typically developing control group; however, this level would be higher than the 18q- levels of conflict.

Three dependent variables were used to test these hypotheses: the Cohesion, Expressiveness, and Conflict subscales of the FES. The independent measure was group (Control, DS, or 18q-). Preliminary assumption testing was conducted. During the preliminary analysis, the variables were found to be considerably skewed, and thus the assumption of normality was violated. According to Stevens (2002), when sample size is equal, MANOVA is robust for violations of normality. Because the 18q- group had only 24 subjects, it was necessary to take a random sample of each of the other two groups in order to create equal groups of 24 subjects each. When the data were analyzed with equal group numbers, a statistically significant difference that was similar to the *F*-value in the original data analysis was revealed, indicating that the MANOVA was in fact robust for the violation of normality. During preliminary analysis, Box's test also revealed a significant finding (p=.026), meaning the assumption of equal variances was violated. According to Mertler and Vannatta (2002), if Box's test is significant, the more robust Pillai's Trace statistic should be used as a test of statistical significance in MANOVA.

Compared to the FES norms, the group means for all three groups were within the average range for the Conflict subscale. The group means for the 18q- and Control groups were in the above average range for the Cohesion subscale. The Control group also demonstrated an above average group mean on the Expressiveness subscale. The DS group and the 18q- demonstrated means within the average range for the Expressiveness subscale, and the DS group also demonstrated a mean in the average range for the Cohesion subscale. Using the Pillai's Trace criterion, the results of the one-way between-groups MANOVA showed a statistically significant multivariate effect for groups on the combined dependent variables, multivariate $F(6,168) = 2.246$, $p = .041$. Analysis of Variance (ANOVA) was conducted on each dependent variable as a follow-up test to MANOVA. In examining the univariate effects of the dependent variables, there are statistically significant differences between the groups on the FES Cohesion subscale, $F(2, 85) = 5.78$, $p = .004$. Differences between groups on the FES Expressiveness subscale were not significant, $F(2, 85) = 2.49$, $p = .089$. Differences between groups on the FES Conflict subscale were also not significant, $F(2, 85) = 2.562$, $p = .083$.

The Tukey HSD post hoc analysis revealed that there are statistically significant differences between the DS group and both the 18q- and the Control groups on the FES Cohesion subscale, with the DS group showing statistically significantly lower levels of cohesion in the family environment than both the 18q- group ($p = .016$) and the Control group ($p = .010$). The post hoc analysis did not reveal any statistically significant differences between the groups on the FES Expressiveness or Conflict subscales.

Although statistical significance was found for the group differences on the Cohesion subscale, it was not in the same direction as predicted in Hypotheses 2a, b, c, and d. Figure 6 demonstrates the expected results of the mean group differences on the three FES subscales. Figure 7 demonstrates the observed results of the mean group differences on the three FES subscales.

Figure 6. *Expected FES Subscale Means*

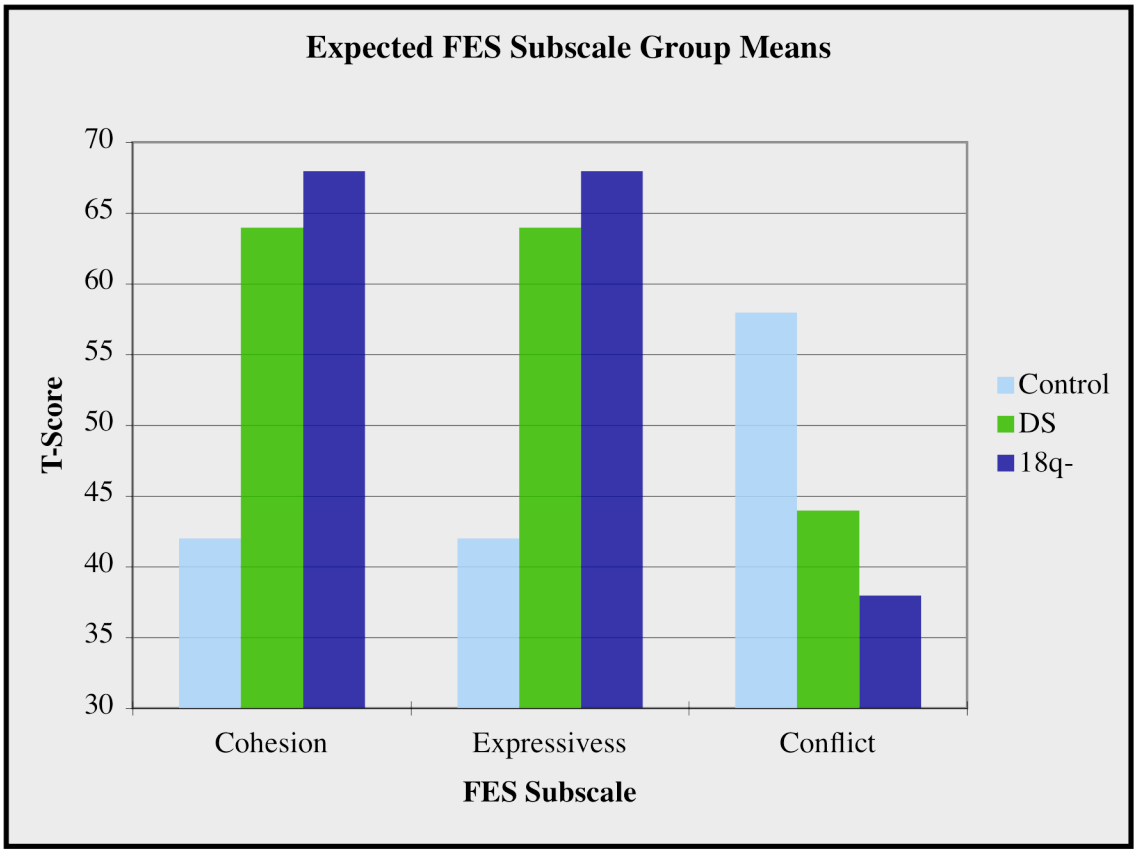
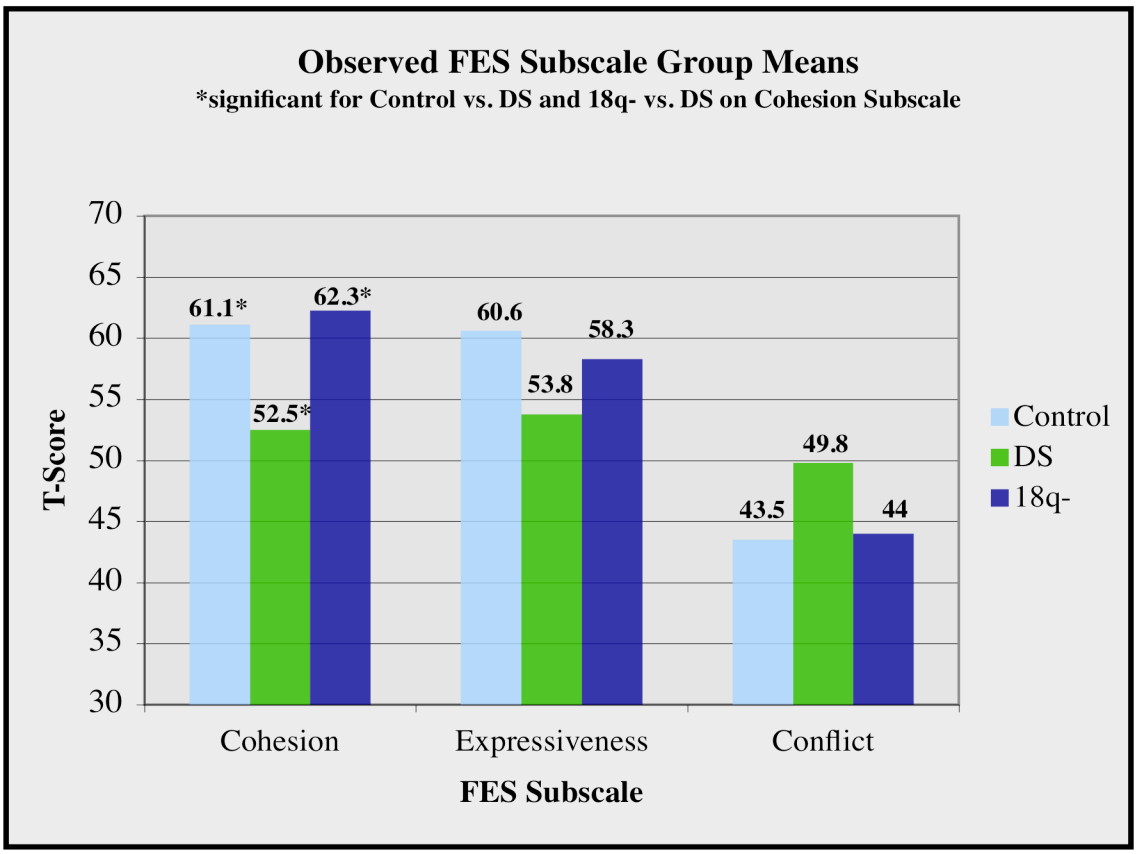


Figure 7. *Observed FES Subscale Means*



Summary

The purpose of the present study was to examine several variables of parenting stress and family environment in families of children with 18q-, Down Syndrome, and typically developing children between the ages of 1 month and 6 years. Additionally, the present research intended to provide a step toward a more in depth understanding of the types of parenting stress and family environment that occur in families of children with and without genetic disorders.

Testing of hypotheses 1a and 1b showed a statistically significant multivariate effect for groups on the combined dependent variables, multivariate $F(6,166) = 2.217$,

p=.044. Further analysis utilizing ANOVA revealed statistically significant differences between the groups on the PSI Isolation subscale, $F(2, 85) = 3.35$, p=.04, and on the PSI Spouse subscale $F(2, 85) = 3.77$, p=.027. A Tukey HSD post hoc analysis was conducted and demonstrated that the DS group reported statistically significantly more stress than the Control group on the PSI Isolation subscale (p=.035). The Tukey HSD analysis also revealed that the DS group reported statistically significantly more stress on the PSI Spouse subscale than the Control group (p=.022). The 18q- group was not found to be statistically significantly different from either the Control group or the DS group on any of the three PSI subscales.

Although statistical significance was found for these two subscales, they were not in the same direction as the hypotheses 1a or 1b expected. Hypothesis 1a predicted that parents of children with 18q- would show above average levels of stress related to competence, isolation, and spouse relationships, and that the families of children with 18q- would show higher levels of stress compared to those with families with DS or typically developing children. It was found, however, that there were no significant differences between the 18q- group and either the DS group or the typically developing group. Hypothesis 1b predicted that parents of children with DS would show above average levels of stress related to competence, isolation, and spouse relationships compared to the PSI norm group and the typically developing control group; however, these levels were expected to be lower than the 18q- levels. It was found, however, that although the DS group showed higher levels of stress related to Isolation and Spouse compared to the 18q- and Control groups, the difference was only statistically significant

for the DS versus Control group. Additionally, the mean scores for all three subscales for the DS group were not in the clinically significant range as compared to the PSI norm groups.

Testing of Hypotheses 2a, 2b, 2c, and 2d showed a statistically significant multivariate effect for groups on the combined dependent variables, multivariate $F(6,168) = 2.246$, $p=.041$. Further analysis utilizing ANOVA revealed statistically significant differences between the groups on the FES Cohesion subscale, $F(2, 85) = 5.78$, $p=.004$. Differences between groups on the FES Expressiveness and FES Conflict subscales were not significant. A Tukey HSD post hoc analysis was conducted and demonstrated that the DS group reported statistically significantly less amounts of Cohesion in the family environment than both the 18q- group ($p=.016$) and the Control group ($p=.010$). The post hoc analysis did not reveal any statistically significant differences between the groups on the FES Expressiveness or Conflict subscales.

Hypothesis 2a predicted that parents of children with 18q- would show above average levels of cohesion and expressiveness in their family environments. The families of children with 18q- were also expected to show higher levels of cohesion and expressiveness compared to those with families with DS or typically developing children. Hypothesis 2c predicted that parents of children with DS would show above average levels of cohesion and expressiveness in their family environments compared to the FES norm group and the typically developing control group; however, these levels will be lower than the 18q- levels of cohesion and expressiveness. It was found, however, that both the 18q- group and the Control group had statistically significantly higher levels of

cohesion in the family environment than the DS group. Both the 18q- and the Control groups' mean scores on the FES Cohesion subscale were also in the above average range compared to the FES norm group.

CHAPTER 5: DISCUSSION

The results and implications of the present study are discussed in detail in this chapter. The chapter begins with a presentation of the results of the study organized by hypothesis. Findings are discussed in the context of past literature. Next, implications for research, methodology, and practice are discussed. Finally, limitations of the present study and directions for future research are presented.

Summary of Results

Hypothesis 1a and 1b

Hypothesis 1a of the present study predicted that primary caregivers of children with 18q- would show above average levels of stress related to competence, isolation, and spousal relationships, and that the families of children with 18q- would show higher levels of stress compared to those with families with DS or typically developing children. The second part of this first hypothesis (Hypothesis 1b) predicted that the primary caregivers of children with DS would show above average levels of stress related to competence, isolation, and spouse relationships compared to the PSI norm group and the typically developing control group; however, these levels were expected to be lower than the 18q- levels. In other words, a continuum of stress was expected for each of the three subscales, with the control group at the lower end of the scale compared to the other groups and within the average range, the DS group in the middle in the above average range, and the 18q- group at the higher end of the scale, also in the above average range.

Children with 18q- experience a multitude of difficulties, including medical complications, behavior problems, delayed cognitive abilities, and below average adaptive skills. It has been shown that stress in the family is elevated when children have more severe developmental and neurological problems (Beckman, 1991; Crnic, Friedrich, & Greenberg, 1983; Dyson, 1996). Because children with Down Syndrome also often exhibit medical complications, impaired cognitive abilities, and other developmental delays, it is expected that levels of parenting stress in these families will be similar to the families of children with 18q-. The DS group was expected to experience less stress than the 18q- group based on the much higher prevalence of DS and the greater availability of resources available to DS families.

The results of these hypotheses were partially confirmed. Statistically significant differences between the DS group and the Control group on two of the three PSI subscales were found, but not for the 18q- group. The 18q- group was not found to be statistically significantly different from either the Control group or the DS group on any of the three PSI subscales. The primary caregivers of the 18q- children scored in the middle range for the Isolation and Spouse subscales (between the Control and the DS participants). They demonstrated the least amount of stress among the three groups on the PSI Competence scale, although the differences between group scores for this subscale were not statistically significant.

The DS group reported statistically significantly higher levels of stress than the Control group on the PSI Isolation subscale, indicating that the primary caregivers in the DS group feel significantly more socially isolated from peers, relatives, and other

emotional support systems than the Control group participants. The analyses also revealed that the DS group reported statistically significantly more stress on the PSI Spouse subscale than the Control group, indicating that the DS primary caregivers experience significantly more spouse-related stress than the Control group participants.

A study by Spangenberg and Theron (2001) revealed that almost one quarter of parents of children with DS in their study were depressed, and that nearly half of the parents of children with DS experienced above average anxiety levels. Walker, Van Slyke, and Newbrough (1991) found that parents of children with mental retardation obtained higher scores on scales that assess stress related to child caretaking demands as compared with control families of typically developing children. The present study supports these findings, demonstrating that the primary caregivers of children with DS exhibited statistically significantly more stress related to the spouse relationship and social isolation than the control group; however, it is unclear why the DS group experienced significantly more stress compared to the 18q- group on these subscales.

In terms of social isolation, Kazak and Marvin (1984) reported that the development and maintenance of friendships is based on sharing common interests and activities, which is difficult for families with handicapped children based on the special demands that these children place on the family. Minnes (1988) found that social support from extended family, friends, and neighbors was negatively correlated with the stress of mothers of children with mental retardation. Similarly, Beckman (1991) found that for both mothers and fathers of children with disabilities, increased informal support was significantly associated with decreased stress. Additional research also demonstrates a

relationship between informal social support and decreases in parenting stress in families of children with disabilities (Floyd & Phillipe, 1993; Kazak & Marvin, 1984). Several studies reveal that families with a handicapped child experience social isolation, and that some parents report feeling that their relationships with friends and family were adversely affected by the birth of a handicapped child (Gayton, 1975; McAndrew, 1976; Waisbren, 1980).

One notable study by Williams, Elder, and Griggs (1987) indicated that of families of children with developmental disabilities, 27% reported that they lacked a support system within the family, while 33% reported that they lacked a support system outside the family such as relatives and friends. The present findings somewhat support the findings by Williams et al. (1987). The DS group reported statistically significantly more stress related to social isolation than the control group, although the 18q- group did not report significantly different levels of social isolation compared to either the Control or the DS group.

It is unclear why the DS group experienced more stress compared to the 18q- group in the area of social isolation. Because DS is a much more prevalent disorder than 18q-, with local agencies in many cities, the DS group was expected to have more opportunities for social interaction with other families of children with DS, more resources for social support, and thus less social isolation than the 18q- group, whose disorder is both less prevalent and less understood. Some studies have indicated that parents of children with mild or marginal handicaps may have more difficulty accepting their children's limitations (Benson and Gross, 1989). Although DS individuals are

generally more moderately impaired (as opposed to mild or marginal), it is possible that the prevalence of DS makes it seem to be more prevalent to the affected parents and possibly involves feelings of responsibility for the child's disability. Additionally, the severe medical complications that accompany the more rare 18q- may allow these parents view their children's limitations in the context of medical fragility rather than mental retardation, even though both disorders entail both cognitive and physical difficulties. It is possible that the many medical difficulties that accompany 18q- make it a diagnosis that is somehow more sympathetic to others and more acceptable to parents than DS. DS has been associated with varied stereotypes and judgments that mistakenly place the responsibility for the disability on the age of the parent. In contrast for the parents of children with 18q-, the chromosomal abnormality is a de novo occurrence for which little responsibility is attributed to the parent. Parents that have difficulty accepting their children's limitations would logically have more difficulty forming and maintaining social relationships; thus, if DS parents have more difficulty accepting their children's diagnosis than parents of children with 18q-, their elevated stress related to both the spousal relationship and social isolation is understandable. These findings are important for interventions that are developed for children with all types of cognitive delays and particularly for parents of children with DS.

In terms of the spousal relationship, Crnic, Friedrich, and Greenberg (1983) noted in their review of the research that studies of marital satisfaction do not reveal a uniform and consistent pattern, and that marital response is likely dependent upon factors aside from the presence of a child with mental retardation, such as severity of the handicap, age

and sex of the child, and the quality of the marital relationship prior to the birth of the child. Benson and Gross (1989) also indicated that the majority of studies regarding marital relationships in families with handicapped children are inconclusive.

The present study provided some insight into the understanding of marital relationships within families of children with disabled children, but also raised some questions: although the DS group did show increased levels of stress on the Spouse subscale, the 18q- group did not show significantly increased levels of stress despite the presence of a handicapped child. It is again uncertain why the DS group experienced more stress compared to the 18q- group in this area; however, it is possible that the severity of the handicap, as well as quality of the marital relationship prior to the child's birth, could play a role. Benson and Gross (1989) suggested that those parents of children with mild or marginal handicaps may have more difficulty accepting their children's limitations. Although direct measure of the marital relationship was beyond the scope of this study, such assessment may be very beneficial when structuring interventions for children with cognitive delays.

Because of the prevalence of DS and the fact that so many individuals with DS are seen in the community (such as working at a store), DS may be viewed as a less severe disability by the public, as well as by parents; therefore, even though many children with DS in fact have moderate difficulties (as opposed to mild or marginal), parents and others may interpret their children's difficulties as less severe than they actually are given the prevalence and public knowledge of DS. The interpretation of DS as less severe than it actually is not only supports Benson and Gross' (1989) findings

regarding parental acceptability of their children, but also demonstrates how a family might experience increased social isolation as well as increased difficulty within the marital relationship.

It should be noted that despite the significant differences demonstrated between groups on two of the PSI subscales, none of the group mean scores for the parenting stress subscales were clinically meaningful; thus, although the DS group experienced significantly higher amounts of stress than the 18q- and Control group on two subscales, the levels of stress that were experienced by the DS group as a whole do not appear to be significantly maladaptive. Although the levels of stress in these families may present as subclinical, it is still likely that these stressors are affecting the parents' own well-being, as well as the parental relationships with their children. Despite the subclinical levels of stress revealed in the present study, the implications of parenting stress remain important in working with families to understand their children's limitations, and to help families cope with the stress related to parenting a child with disabilities.

Hypothesis 2a, 2b, 2c, and 2d

Hypothesis 2a of the present study predicted that primary caregivers of children with 18q- would show above average levels of cohesion and expressiveness in their family environments. The families of children with 18q- were also expected to show higher levels of cohesion and expressiveness compared to those with families with DS or typically developing children. Hypothesis 2b predicted that primary caregivers of children with 18q- would show below average levels of conflict in their family environments, and that the families of children with 18q- would show lower levels of

conflict compared to those of families with DS or typically developing children. Hypothesis 2c predicted that parents of children with DS would show above average levels of cohesion and expressiveness in their family environments compared to the FES norm group and the typically developing control group; however, these levels will be lower than the 18q- levels of cohesion and expressiveness. Hypothesis 2d predicted that parents of children with DS would show below average levels of conflict in their family environments compared to the FES norm group and the typically developing control group; however, this level would be higher than the 18q- levels of conflict.

In other words, two continuums were expected for these FES subscales. In the first continuum, the 18q- group was expected to experience the greatest amount of cohesion and expressiveness in the family environment in the above average range. The DS group was expected to demonstrate scores between the 18q- and the Control group for cohesion and expressiveness, but also in the above average range. The control group was expected to be at the lowest end of the continuum and within the average range for cohesion and expressiveness. In the second continuum, the Control group was expected to have the highest levels of conflict in the family environment, followed by the DS group with the middle level of conflict in the below average range, and the 18q- group with the lowest amount of conflict, also in the below average range. Based on this hypothesis, the DS group was expected to experience less cohesion and expressiveness and more conflict than the 18q- group.

These hypotheses were partially confirmed. Results of the tests of these hypotheses revealed a statistically significant difference between the DS group and both

the 18q- and the Control groups on the FES Cohesion subscale, with the DS group showing statistically significantly less amounts of cohesion in the family environment than both the 18q- group and the Control group, which was contrary to the expectations. It is interesting to note that the Control and 18q- groups in fact showed above average levels of Cohesion compared to the FES norm group, while the DS group reported significantly less Cohesion than both groups. Given the DS group's higher levels of stress on the two subscales of the PSI in the previous section of this study, it is logical to deduce that families with higher stress levels related to the spouse relationship and social isolation would report lower levels of cohesiveness within the family environment. Although the DS group reported significantly lower levels of cohesion in the family environment than both the 18q- and the Control groups, the mean score for the DS group on the Cohesion subscale was still within the average range and does not appear to be significantly maladaptive.

On the Expressiveness subscale, the Control group exhibited the greatest amount of Expressiveness in the family environment, followed by the 18q- and the DS group, although the differences between group scores for Expressiveness subscale scores were not statistically significant. On the Conflict subscale, the DS group exhibited the highest amount of Conflict in the family environment, followed by the 18q- and Control groups; however, the differences between group scores for the Conflict subscale scores were not statistically significant. The Control group's mean scores on both Cohesion and Expressiveness were in the above average range, and the 18q- group's mean score for

Cohesion was in the above average range. The other mean scores for each group on each of the three FES subscales were within the average range.

Several studies have concluded that although parenting stress may increase when families must cope with a disabled child, the basic dimensions of family functioning are not necessarily disrupted (Cadman, Rosenbaum, Boyle, & Offord, 1991; Kazak, 1987; Walker, Van Slyke, & Newbrough, 1992). Dyson (1991) noted that although stress in families of handicapped children is elevated, this stress does not appear to be predictive of family dysfunction. Dyson concluded that families generally appear to respond to the care of a handicapped child with resilience and adaptive functioning despite the presence of family stress. Aside from Dyson's conclusions, families do exist that are not resilient and have difficulty adapting to changes. Some factors that may limit a family's ability to cope include martial discord, marital status, and financial difficulties. The socioeconomic status of the participants in the present study is unknown; however, 88.6% of primary caregivers were married at the time of the survey.

In their study of families of children with Down Syndrome, Van Riper, Ryff, and Pridham (1992) found that families with a child with Down Syndrome are more comparable to than different from families of nondisabled children, and Mahoney and O'Sullivan (1992) concluded that in general, the interpersonal relationships as measured by cohesion, conflict, and expressiveness appeared slightly more favorable for families of children with disabilities than for the normative sample of families. Pueschel and Myers (1994) studied the family environments of children with DS and found that high scores on the FES were observed on the Cohesion and Expressiveness subscales, among others.

They concluded that these scores indicated a high degree of commitment and support family members provide for one another as well as the extent to which members are encouraged to express their feelings. Furthermore, these studies indicated that families with handicapped children may experience a greater need to become more cohesive and supportive in the face of these difficulties. Based on these findings, as well as the fact that children with 18q- and DS exhibit similar severe impairments, the DS and 18q- groups were expected to exhibit above average levels of cohesion and expressiveness and below average levels of conflict as compared to the FES norm group and the typically developing control group. In the present study, however, the DS group mean scores were in the average range for all three subscales. Although the 18q- group had a mean score in the above average range on the Cohesion subscale as expected, the 18q- group mean score on the Expressiveness subscale was in the average range. The Control group of families of typically developing children was expected to be in the average range for all subscales; however, they scored in the above average range on both the Cohesion and Expressiveness subscales.

It should be noted that not all studies have found higher amounts of expressiveness and cohesion in families of handicapped children. A study by Margalit and Raviv (1983) found that mothers of children with mental retardation viewed their families as less encouraging of open expression of emotions. Another study by Margalit and Heiman (1986) found a decrease in the expression of emotion and the cohesiveness of the family system as a whole related to the presence of a learning disabled child. Blacher, Nihira, and Meyers (1987) found that families of children with severe mental

retardation had the lowest scores on all of the subscales compared to control groups of children with mild and moderate mental retardation. Given the earlier publication dates of these studies, however, the present study expected similar results to the more current research as mentioned previously, which revealed positive family environments in families of disabled children.

As discussed in the results for Hypothesis 1, although the family environments may present as average compared to the FES norm group, it is still possible that subtle differences in family environments affect not only well-being of the individuals in the family, but also the relationships between the family members. Despite the average levels of conflict within the groups, family environment remains an important factor in working with families to understand their children's limitations, and to help families cope with the stress related to parenting a child with disabilities.

Implications of Findings

Findings from the present study have implications for support services and daily functioning for families coping with an initial diagnosis of a disability or genetic disorder, as well as for families that already have a child with a disability and are experiencing difficulties related to family functioning and stress. The results of this study are particularly helpful for mental health professionals working with families of children with disabilities. By becoming aware of the effects of a child's disability on parenting stress and the family environment, mental health professionals can help families create and maintain more supportive, healthy relationships in the context of coping with a child's limitations.

An important implication of this study is that all children with mental retardation are not the same, and likewise families of disabled children differ in their coping strategies, approaches to diagnoses, and management of stress and difficulty. These differences are especially relevant in clinical settings, where practitioners may categorize and treat clients based on common characteristics. It is important that practitioners recognize the importance of the family system in working with children with genetic disorders. Development of treatments and interventions for children with disabilities must not only be specific to the child's limitations, but also to the family's understanding of the child's abilities. Awareness of family attributes, such as stress levels and types of environment, is vital both in developing appropriate interventions for the child as well as in gaining parental support for the implementation of such interventions.

Given the findings of the present study, it is important that both medical and mental health professionals work with families to understand their children's limitations, and to help families cope with the stress related to parenting a child with disabilities. It is evident from this study that stress related to parenting a disabled child is increased in the areas of the spousal relationship and feelings of social isolation. These findings should guide clinicians in terms of what areas to be aware of when working with these families. For instance, clinicians can assist parents in finding social support or facilitate their understanding of the effects of parenting a handicapped child on the spousal relationship.

This study demonstrated statistically significant correlations among all of the subscales. This is an important finding, because it allows both researchers and clinicians to recognize that many aspects of family functioning are related. Looking at separate

variables helped determine what specific areas of functioning were statistically significantly different between the groups, but the recognition of the interrelatedness of the 6 subscales provides a greater frame of reference for examining family attributes.

Limitations and Future Directions

Several limitations of the present study are related to the sample of primary caregivers recruited to participate in this study. Because convenience sampling methods were utilized, this sample may be made up of a more homogenous group of individuals than the actual population of primary caregivers of children with disabilities. Individuals who volunteered to participate in this study in the DS or 18q- groups were involved to some extent with a research or community organization. This involvement and decision to volunteer indicates some degree of awareness and commitment that others in the population may not possess. The sample used in this study was also drawn from a limited geographical area, as well as possibly from a limited socioeconomic and cultural stratum. These factors, in combination with the small sample size, mean that the results of this study cannot be generalized across geographic areas, cultures, or socioeconomic levels.

Another limitation of this study involves the use of self-report measures, which may be less reliable when completed outside of a clinical setting. These self-report measures are also fairly transparent as far as what is being asked, and the parent may either fake good or bad. Without a validity scale on these measures, it is difficult to know how accurate they reflect the family unit. Thus, because this study dealt with personal issues related to marriage, parenting, and family, it is possible that the

participants were not truthful in their responses. It is important to consider how the individual characteristics and response styles of the participants may have contributed to the findings of the present study.

Future research in this field should recruit a larger, broader, random sample of primary caregivers across a more evenly distributed range of ethnicity, socioeconomic status, and geographical location. There are several other areas related to the current study that future research should also investigate. Examination of adaptive skills was beyond the scope of this study; however, research that examines the relationship between a child's adaptive skills and parenting stress may reveal a more specific understanding of the influences of a handicapped child on the family. Additionally, future research should examine parenting stress and the family environment within intact versus non-intact families to determine the relationship between having a child with a disability and marital discord or divorce.

Conclusions

The present study investigated several variables of parenting stress and family environment in families of children with and without disabilities. The three groups examined in this study were primary caregivers of children with 18q-, primary caregivers of children with DS, and a control group of primary caregivers of typically developing children. The DS group reported statistically significantly more stress than the Control group on both the Isolation and Spouse subscales of the PSI. The 18q- group was not found to be statistically significantly different from either the Control or DS group on any of the three PSI subscales. The DS group showed statistically significantly less amounts

of cohesion on the FES than both the 18q- and Control groups. The 18q- group showed similar levels of cohesion to the Control group. There were no significant differences between groups on the Expressiveness or Conflict subscales of the FES.

Findings from the present study provide important information about the role of family environment and parenting stress in families of children with disabilities. Both medical and mental health professionals should recognize the importance of the family system in working with children with genetic disorders, especially in terms of developing treatment plans and interventions. These findings should guide clinicians in terms of what areas to be aware of when working with these families, such as the spousal relationship and social support. Further exploration of the associations between adaptive skills, marital status, and family functioning will be helpful in gaining a more specific understanding of the influences of a handicapped child on the family and thus developing more appropriate support services and intervention programs.

APPENDICES

Appendix A: Family Environment Scale

mind garden

Family Environment Scale

Form R

Item Booklet

Rudolf H. Moos

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Instructions

There are 90 statements in this booklet. They are statements about families. You are to decide which of these statements are true of your family and which are false. Make all your marks on the separate answer sheet. If you think the statement is *True* or mostly *True* of your family, make an X in the box labeled T (true). If you think the statement is *False* or mostly *False* of your family, make an X in the box labeled F (false).

You may feel that some of the statements are true for some family members and false for others. Mark T if the statement is *true* for most members. Mark F if the statement is *false* for most members. If the members are evenly divided, decide what is the stronger overall impression and answer accordingly.

Remember, we would like to know what your family seems like to *you*. So do not try to figure out how other members see your family, but *do* give us your general impression of your family for each statement.

Work Across →

1. Family members really help and support one another.
2. Family members often keep their feelings to themselves.
3. We fight a lot in our family.
4. We don't do things on our own very often in our family.
5. We feel it is important to be the best at whatever you do.
6. We often talk about political and social problems.
7. We spend most weekends and evenings at home.
8. Family members attend church, synagogue, or Sunday School fairly often.
9. Activities in our family are pretty carefully planned.
10. Family members are rarely ordered around.
11. We often seem to be killing time at home.
12. We say anything we want to around home.
13. Family members rarely become openly angry.
14. In our family, we are strongly encouraged to be independent.
15. Getting ahead in life is very important in our family.
16. We rarely go to lectures, plays or concerts.
17. Friends often come over for dinner or to visit.
18. We don't say prayers in our family.
19. We are generally very neat and orderly.
20. There are very few rules to follow in our family.
21. We put a lot of energy into what we do at home.
22. It's hard to "blow off steam" at home without upsetting somebody.
23. Family members sometimes get so angry they throw things.
24. We think things out for ourselves in our family.
25. How much money a person makes is not very important to us.
26. Learning about new and different things is very important in our family.
27. Nobody in our family is active in sports, Little League, bowling, etc.
28. We often talk about the religious meaning of Christmas, Passover, or other holidays.
29. It's often hard to find things when you need them in our household.
30. There is one family member who makes most of the decisions.

31. There is a feeling of togetherness in our family.
32. We tell each other about our personal problems.
33. Family member hardly ever lose their tempers.
34. We come and go as we want to in our family.
35. We believe in competition and "may the best man win."
36. We are not that interested in cultural activities.
37. We often go to the movies, sports events, camping, etc.
38. We don't believe in heaven or hell.
39. Being on time is very important in our family.
40. There are set ways of doing things at home.
41. We rarely volunteer when something has to be done at home.
42. If we feel like doing something on the spur of the moment we often just pick up and go.
43. Family members often criticize each other.
44. There is very little privacy in our family.
45. We always strive to do things just a little better the next time.
46. We rarely have intellectual discussions.
47. Everyone in our family has a hobby or two.
48. Family members have strict ideas about what is right and wrong.
49. People change their minds often in our family.
50. There is a strong emphasis on following rules in our family.
51. Family members really back each other up.
52. Someone usually gets upset if you complain in our family.
53. Family members sometimes hit each other.
54. Family members almost always rely on themselves when a problem comes up.
55. Family members rarely worry about job promotions, school grades, etc.
56. Someone in our family plays a musical instrument.
57. Family members are not very involved in recreational activities outside work or school.
58. We believe there are some things you just have to take on faith.
59. Family members make sure their rooms are neat.
60. Everyone has an equal say in family decisions.

61. There is very little group spirit in our family.
62. Money and paying bills is openly talked about in our family.
63. In there's a disagreement in our family, we try hard to smooth things over and keep the peace.
64. Family members strongly encourage each other to stand up for their rights.
65. In our family, we don't try that hard to succeed.
66. Family members often go to the library.
67. Family members sometimes attend courses or take lessons for some hobby or interest (outside of school).
68. In our family each person has different ideas about what is right and wrong.
69. Each person's duties are clearly defined in our family.
70. We can do whatever we want to in our family.
71. We really get along well with each other.
72. We are usually careful about what we say to each other.
73. Family members often try to one-up or out-do each other.
74. It's hard to be by yourself without hurting someone's feelings in our household.
75. "Work before play" is the rule in our family.
76. Watching TV is more important than reading in our family.
77. Family members go out a lot.
78. The Bible is a very important book in our home.
79. Money is not handled very carefully in our family.
80. Rules are pretty inflexible in our household.
81. There is plenty of time and attention for everyone in our family.
82. There are a lot of spontaneous discussions in our family.
83. In our family, we believe you don't ever get anywhere by raising your voice.
84. We are not really encouraged to speak up for ourselves in our family.
85. Family members are often compared with others as to how well they are doing at work or school.
86. Family members really like music, art and literature.
87. Our main form of entertainment is watching TV or listening to the radio.
88. Family members believe that if you sin you will be punished.
89. Dishes are usually done immediately after eating.
90. You can't get away with much in our family.

Family Environment Scale

Answer Sheet

Look at your Family Environment Scale item booklet and check the Form printed on it here:

Form R___ I___ E___

Please provide the information requested below.

Your Name_____ Age_____

Address _____ Gender: Male Female
(circle)

Please indicate your position in the family (check one):

Mother (wife)____ Father (husband)_____ Son or Daughter____

Other_____ (Please Specify):_____

Today's Date_____

Now, please read the instructions on the front page of your Family Environment Scale Item booklet and be sure that you understand them. When you are ready, read each statement in your booklet and then, in the boxes on page 2 of this sheet, mark T (true) if you think the statement is true of your family, and F (false) if the statement is not true of your family.

Use a heavy X, as in the example: Please use a pencil with an eraser, not a pen. Be sure to match each number in the booklet with each one on this sheet.

T	X	
	1	2
F		X

START HERE

T/F											T/F
T/F	1	2	3	4	5	6	7	8	9	10	T/F
T/F	11	12	13	14	15	16	17	18	19	20	T/F
T/F	21	22	23	24	25	26	27	28	29	30	T/F
T/F	31	32	33	34	35	36	37	38	39	40	T/F
T/F	41	42	43	44	45	46	47	48	49	50	T/F
T/F	51	52	53	54	55	56	57	58	59	60	T/F
T/F	61	62	63	64	65	66	67	68	69	70	T/F
T/F	71	72	73	74	75	76	77	78	79	80	T/F
T/F	81	82	83	84	85	86	87	88	89	90	T/F

---

do not mark below this line

	C	Ex	Con	Ind	AO	ICO	ARO	MRE	Org	Ctl
R/S										
S/S										

Appendix B: Parenting Stress Index

PSI Item Booklet

Instructions:

On the PSI Answer Sheet, please write your name, gender, date of birth, ethnic group, marital status, child's name, child's gender, child's date of birth, and today's date. Please mark all your responses on the answer sheet. DO NOT WRITE ON THIS BOOKLET.

This questionnaire contains 120 statements. Read each statement carefully. For each statement, please focus on the child you are most concerned about, and circle the response which best represents your opinion.

Circle the SA if you strongly agree with the statement.
Circle the A if you agree with the statement.
Circle the NS if you are not sure.
Circle the D if you disagree with the statement.
Circle the SD if you strongly disagree with the statement.

For example, if you sometimes enjoy going to the movies, you would circle A in response to the following statement:

I enjoy going to the movies. SA (A) NS D SD

While you may not find a response that exactly states your feelings, please circle the response that comes closest to describing how you feel. YOUR FIRST REACTION TO EACH QUESTION SHOULD BE YOUR ANSWER.

Circle only one response for each statement, and respond to all statements. DO NOT ERASE! If you need to change an answer, make an "X" through the incorrect answer and circle the correct response. For example:

I enjoy going to the movies. SA A NS (X) (SD)

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1. When my child wants something, my child usually keeps trying to get it.
2. My child is so active that it exhausts me.
3. My child appears disorganized and is easily distracted.
4. Compared to most, my child has more difficulty concentrating and paying attention.
5. My child will often stay occupied with a toy for more than 10 minutes.
6. My child wanders away much more than I expected.
7. My child is much more active than I expected.
8. My child squirms and kicks a great deal when being dressed or bathed.
9. My child can be easily distracted from wanting something.
10. My child rarely does things for me that make me feel good.
11. Most times I feel that my child likes me and wants to be close to me.
12. Sometimes I feel my child doesn't like me and doesn't want to be close to me.
13. My child smiles at me much less than I expected.
14. When I do things for my child, I get the feeling that my efforts are not appreciated very much.

For statement 15, choose a response from choices 1 to 4 below.

15. Which statement best describes your child?
    1. almost always likes to play with me
    2. sometimes likes to play with me
    3. usually doesn't like to play with me
    4. almost never likes to play with me

For statement 16, choose a response from choices 1 to 5 below.

16. My child cries and fusses:
    1. much less than I had expected
    2. less than I expected
    3. about as much as I expected
    4. much more than I expected
    5. it seems almost constant
17. My child seems to cry or fuss more often than most children.
18. When playing, my child doesn't often giggle or laugh.
19. My child generally wakes up in a bad mood.
20. I feel that my child is very moody and easily upset.
21. My child looks a little different than I expected and it bothers me at times.
22. In some areas, my child seems to have forgotten past learnings and has gone back to doing things characteristic of younger children.
23. My child doesn't seem to learn as quickly as most children.
24. My child doesn't seem to smile as much as most children.

2

25. My child does a few things which bother me a great deal.

26. My child is not able to do as much as I expected.

27. My child does not like to be cuddled or touched very much.

28. When my child came home from the hospital, I had doubtful feelings about my ability to handle being a parent.

29. Being a parent is harder than I thought it would be.

30. I feel capable and on top of things when I am caring for my child.

31. Compared to the average child, my child has a great deal of difficulty in getting used to changes in schedules or changes around the house.

32. My child reacts very strongly when something happens that my child doesn't like.

33. Leaving my child with a babysitter is usually a problem.

34. My child gets upset easily over the smallest thing.

35. My child easily notices and overreacts to loud sounds and bright lights.

36. My child's sleeping or eating schedule was much harder to establish than I expected.

37. My child usually avoids a new toy for a while before beginning to play with it.

38. It takes a long time and it is very hard for my child to get used to new things.

39. My child doesn't seem comfortable when meeting strangers.

For statement 40, choose from choices 1 to 4 below.

40. When upset, my child is:
    1. easy to calm down
    2. harder to calm down than I expected
    3. very difficult to calm down
    4. nothing I do helps to calm my child

For statement 41, choose from choices 1 to 5 below.

41. I have found that getting my child to do something or stop doing something is:
    1. much harder than I expected
    2. somewhat harder than I expected
    3. about as hard as I expected
    4. somewhat easier than I expected
    5. much easier than I expected

For statement 42, choose from choices 1 to 5 below.

42. Think carefully and count the number of things which your child does that bothers you. For example: dawdles, refuses to listen, overactive, cries, interrupts, fights, whines, etc. Please circle the number which includes the number of things you counted.
    1. 1–3
    2. 4–5
    3. 6–7
    4. 8–9
    5. 10+

For statement 43, choose from choices 1 to 5 below.

43. When my child cries, it usually lasts:
    1. less than 2 minutes
    2. 2–5 minutes
    3. 5 10 minutes
    4. 10–15 minutes
    5. more than 15 minutes

44. There are some things my child does that really bother me a lot.

45. My child has had more health problems than I expected.

46. As my child has grown older and become more independent, I find myself more worried that my child will get hurt or into trouble.

47. My child turned out to be more of a problem than I had expected.

48. My child seems to be much harder to care for than most.

49. My child is always hanging on me.

50. My child makes more demands on me than most children.

51. I can't make decisions without help.

52. I have had many more problems raising children than I expected.

53. I enjoy being a parent.

54. I feel that I am successful most of the time when I try to get my child to do or not do something.

55. Since I brought my last child home from the hospital, I find that I am not able to take care of this child as well as I thought I could. I need help.

56. I often have the feeling that I cannot handle things very well.

For statement 57, choose from choices 1 to 5 below.

57. When I think about myself as a parent I believe:
    1. I can handle anything that happens
    2. I can handle most things pretty well
    3. sometimes I have doubts, but find that I handle most things without any problems
    4. I have some doubts about being able to handle things
    5. I don't think I handle things very well at all

For statement 58, choose from choices 1 to 5 below.

58. I feel that I am:
    1. a very good parent
    2. a better than average parent
    3. an average parent
    4. a person who has some trouble being a parent
    5. not very good at being a parent

For questions 59 and 60, choose from choices 1 to 5 below.

59. What were the highest levels in school or college you and the child's father/mother have completed?
Mother:
    1. 1st to 8th grade
    2. 9th to 12th grade
    3. vocational or some college
    4. college graduate
    5. graduate or professional school

60. Father:
    1. 1st to 8th grade
    2. 9th to 12th grade
    3. vocational or some college
    4. college graduate
    5. graduate or professional school

For question 61, choose from choices 1 to 5 below.

61. How easy is it for you to understand what your child wants or needs?
    1. very easy
    2. easy
    3. somewhat difficult
    4. it is very hard
    5. I usually can't figure out what the problem is

62. It takes a long time for parents to develop close, warm feelings for their children.

63. I expected to have closer and warmer feelings for my child than I do and this bothers me.

64. Sometimes my child does things that bother me just to be mean.

65. When I was young, I never felt comfortable holding or taking care of children.

66. My child knows I am his or her parent and wants me more than other people.

67. The number of children that I have now is too many.

68. Most of my life is spent doing things for my child.

69. I find myself giving up more of my life to meet my children's needs than I ever expected.

70. I feel trapped by my responsibilities as a parent.

71. I often feel that my child's needs control my life.

72. Since having this child, I have been unable to do new and different things.

73. Since having a child, I feel that I am almost never able to do things that I like to do.

74. It is hard to find a place in our home where I can go to be by myself.

75. When I think about the kind of parent I am, I often feel guilty or bad about myself.

76. I am unhappy with the last purchase of clothing I made for myself.

77. When my child misbehaves or fusses too much, I feel responsible, as if I didn't do something right.

78. I feel every time my child does something wrong, it is really my fault.

79. I often feel guilty about the way I feel toward my child.

80. There are quite a few things that bother me about my life.

81. I felt sadder and more depressed than I expected after leaving the hospital with my baby.

82. I wind up feeling guilty when I get angry at my child and this bothers me.

83. After my child had been home from the hospital for about a month, I noticed that I was feeling more sad and depressed than I had expected.

84. Since having my child, my spouse (or male/female friend) has not given me as much help and support as I expected.

85. Having a child has caused more problems than I expected in my relationship with my spouse (or male/female friend).

86. Since having a child, my spouse (or male/female friend) and I don't do as many things together.

87. Since having a child, my spouse (or male/female friend) and I don't spend as much time together as a family as I had expected.

88. Since having my last child, I have had less interest in sex.

89. Having a child seems to have increased the number of problems we have with in-laws and relatives.

90. Having children has been much more expensive than I had expected.

91. I feel alone and without friends.

92. When I go to a party, I usually expect not to enjoy myself.

93. I am not as interested in people as I used to be.

94. I often have the feeling that other people my own age don't particularly like my company.

95. When I run into a problem taking care of my children, I have a lot of people to whom I can talk to get help or advice.

96. Since having children, I have a lot fewer chances to see my friends and to make new friends.

97. During the past six months, I have been sicker than usual or have had more aches and pains than I normally do.

98. Physically, I feel good most of the time.

99. Having a child has caused changes in the way I sleep.

100. I don't enjoy things as I used to.

For statement 101, choose from choices 1 to 4 below.

101. Since I've had my child:
    1. I have been sick a great deal
    2. I haven't felt as good
    3. I haven't noticed any change in my health
    4. I have been healthier

For statements 102 to 120, choose from choices Y for "Yes" and N for "No."
During the last 12 months, have any of the following events occurred in your immediate family?

102. Divorce
103. Marital reconciliation
104. Marriage
105. Separation
106. Pregnancy
107. Other relative moved into household
108. Income increased substantially (20% or more)
109. Went deeply into debt
110. Moved to new location
111. Promotion at work
112. Income decreased substantially
113. Alcohol or drug problem
114. Death of close family friend
115. Began new job
116. Entered new school
117. Trouble with superiors at work
118. Trouble with teachers at school
119. Legal problems
120. Death of immediate family member

PSI Answer Sheet

Name _____ Gender _____ Date of birth _____ Ethnic group _____

Marital status _____ Child's name _____ Child's gender _____

Child's date of birth _____ Today's date _____

SA = Strongly Agree A = Agree NS = Not Sure D = Disagree SD = Strongly Disagree

1. SA A NS D SD	31. SA A NS D SD	61. 1 2 3 4 5	91. SA A NS D SD
2. SA A NS D SD	32. SA A NS D SD	62. SA A NS D SD	92. SA A NS D SD
3. SA A NS D SD	33. SA A NS D SD	63. SA A NS D SD	93. SA A NS D SD
4. SA A NS D SD	34. SA A NS D SD	64. SA A NS D SD	94. SA A NS D SD
5. SA A NS D SD	35. SA A NS D SD	65. SA A NS D SD	95. SA A NS D SD
6. SA A NS D SD	36. SA A NS D SD	66. SA A NS D SD	96. SA A NS D SD
7. SA A NS D SD	37. SA A NS D SD	67. SA A NS D SD	97. SA A NS D SD
8. SA A NS D SD	38. SA A NS D SD	68. SA A NS D SD	98. SA A NS D SD
9. SA A NS D SD	39. SA A NS D SD	69. SA A NS D SD	99. SA A NS D SD
10. SA A NS D SD	40. 1 2 3 4	70. SA A NS D SD	100. SA A NS D SD
11. SA A NS D SD	41. 1 2 3 4 5	71. SA A NS D SD	101. 1 2 3 4
12. SA A NS D SD	42. 1 2 3 4 5	72. SA A NS D SD	102. Y N
13. SA A NS D SD	43. 1 2 3 4 5	73. SA A NS D SD	103. Y N
14. SA A NS D SD	44. SA A NS D SD	74. SA A NS D SD	104. Y N
15. 1 2 3 4	45. SA A NS D SD	75. SA A NS D SD	105. Y N
16. 1 2 3 4 5	46. SA A NS D SD	76. SA A NS D SD	106. Y N
17. SA A NS D SD	47. SA A NS D SD	77. SA A NS D SD	107. Y N
18. SA A NS D SD	48. SA A NS D SD	78. SA A NS D SD	108. Y N
19. SA A NS D SD	49. SA A NS D SD	79. SA A NS D SD	109. Y N
20. SA A NS D SD	50. SA A NS D SD	80. SA A NS D SD	110. Y N
21. SA A NS D SD	51. SA A NS D SD	81. SA A NS D SD	111. Y N
22. SA A NS D SD	52. SA A NS D SD	82. SA A NS D SD	112. Y N
23. SA A NS D SD	53. SA A NS D SD	83. SA A NS D SD	113. Y N
24. SA A NS D SD	54. SA A NS D SD	84. SA A NS D SD	114. Y N
25. SA A NS D SD	55. SA A NS D SD	85. SA A NS D SD	115. Y N
26. SA A NS D SD	56. SA A NS D SD	86. SA A NS D SD	116. Y N
27. SA A NS D SD	57. 1 2 3 4 5	87. SA A NS D SD	117. Y N
28. SA A NS D SD	58. 1 2 3 4 5	88. SA A NS D SD	118. Y N
29. SA A NS D SD	59. 1 2 3 4 5	89. SA A NS D SD	119. Y N
30. SA A NS D SD	60. 1 2 3 4 5	90. SA A NS D SD	120. Y N

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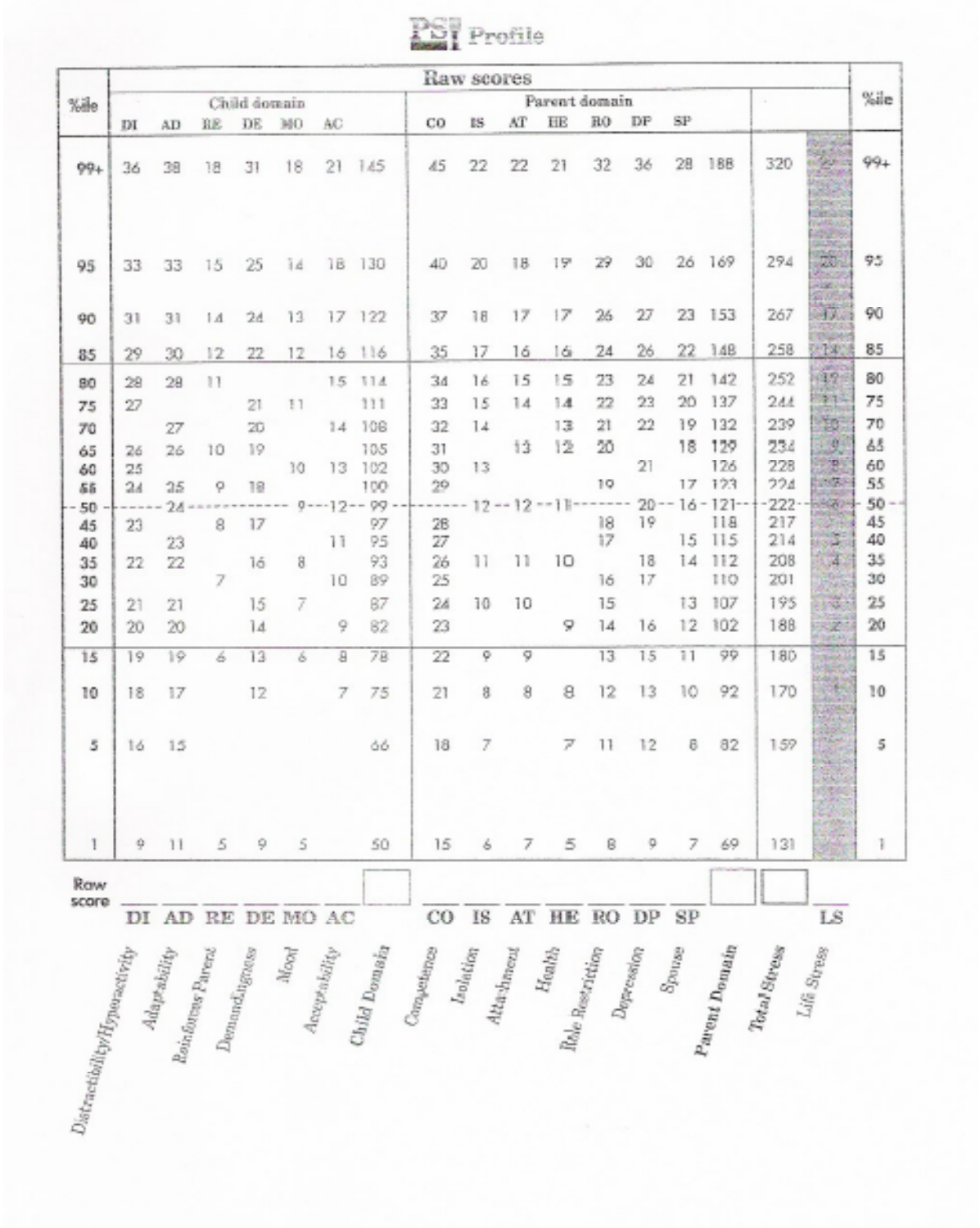
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95 33 33 15 25 14 18 130 40 20 18 19 29 30 26 169 294 95
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80 28 28 11 15 114 34 16 15 15 23 24 21 142 252 80
75 27 21 11 111 33 15 14 14 22 23 20 137 244 75
70 27 20 14 108 32 14 13 21 22 19 132 239 70
65 26 26 10 19 105 31 13 12 20 18 129 234 65
60 25 10 13 102 30 13 21 126 228 60
55 24 25 9 18 100 29 19 17 123 224 55
50 24 9 12 99 12 12 11 20 16 121 222 50
45 23 8 17 97 28 18 19 118 217 45
40 27 23 11 95 27 17 15 115 214 40
35 22 22 16 8 93 26 11 11 10 18 14 112 208 35
30 7 10 89 25 16 17 110 201 30
25 21 21 15 7 87 24 10 10 15 13 107 195 25
20 20 20 14 9 82 23 9 14 16 12 102 188 20
15 19 19 6 13 6 8 78 22 9 9 13 15 11 99 180 15
10 18 17 12 7 75 21 8 8 8 12 13 10 92 170 10
5 16 15 66 18 7 7 11 12 8 82 159 5
1 9 11 5 9 5 50 15 6 7 5 8 9 7 69 131 1
Raw score
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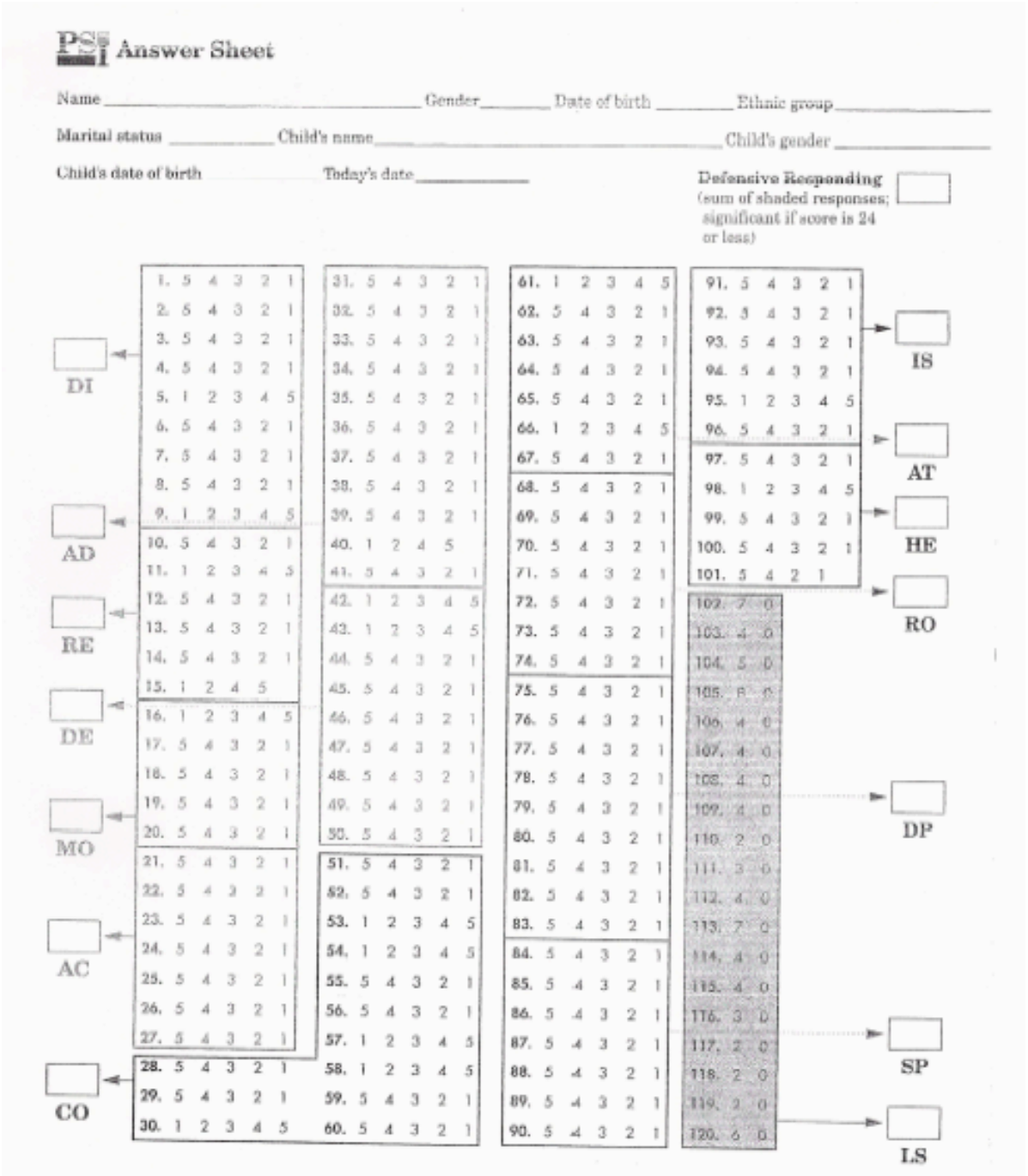
Distractibility/Hyperactivity
Adaptability
Reinforces Parent
Demandingness
Mood
Acceptability
Child Domain
Competence
Isolation
Attachment
Health
Role Restriction
Depression
Spouse
Parent Domain
Total Stress
Life Stress


PSI Answer Sheet
Name
Gender
Date of birth
Ethnic group
Marital status
Child's name
Child's gender
Child's date of birth
Today's date
Defensive Responding
(sum of shaded responses; significant if score is 24 or less)
DI
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1. 5 4 3 2 1
2. 5 4 3 2 1
3. 5 4 3 2 1
4. 5 4 3 2 1
5. 1 2 3 4 5
6. 5 4 3 2 1
7. 5 4 3 2 1
8. 5 4 3 2 1
9. 1 2 3 4 5
10. 5 4 3 2 1
11. 1 2 3 4 5
12. 5 4 3 2 1
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30. 1 2 3 4 5
31. 5 4 3 2 1
32. 5 4 3 2 1
33. 5 4 3 2 1
34. 5 4 3 2 1
35. 5 4 3 2 1
36. 5 4 3 2 1
37. 5 4 3 2 1
38. 5 4 3 2 1
39. 5 4 3 2 1
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42. 1 2 3 4 5
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55. 5 4 3 2 1
56. 5 4 3 2 1
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71. 5 4 3 2 1
72. 5 4 3 2 1
73. 5 4 3 2 1
74. 5 4 3 2 1
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97. 5 4 3 2 1
98. 1 2 3 4 5
99. 5 4 3 2 1
100. 5 4 3 2 1
101. 5 4 2 1
102. 7 0
103. 4 0
104. 5 0
105. 5 0
106. 4 0
107. 4 0
108. 4 0
109. 4 0
110. 2 0
111. 3 0
112. 4 0
113. 7 0
114. 4 0
115. 4 0
116. 3 0
117. 2 0
118. 2 0
119. 2 0
120. 6 0
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Appendix C: Referral Letter
18q- Participants

The University of Texas
Health Science Center at San Antonio
Mail Code 7809
7703 Floyd Curl Drive
San Antonio, Texas 78229-3900

Dear Families,

The Chromosome 18 Clinical Research Center would like for you to participate in a developmental mail out survey on behalf of your child on the topic of parenting stress and family environment. Kim Davis, our neuropsychology consultant from UT-Austin, is compiling data for her dissertation on parenting stress and family environment. Kim's study will examine the influences of genetic disorders such as 18q- and Down Syndrome on various aspects of family functioning.

The extensive impairments that are often associated with 18q-, both physical and behavioral, may be a significant source of stress to parents. Previous research that has focused on stress related to parenting children with mental retardation, genetic disorders, and pervasive developmental disorders reveals high amounts of anxiety regarding both the maladaptive behavior of these children, as well as the effects of such disorders on the family's environment and relationships. To date, there have been no studies that have evaluated the family variables in the 18q- disorder.

Enclosed you will find a copy of the Parenting Stress Index (PSI) and the Family Environment Scale (FES). The PSI is a self-report measure that provides an estimate of areas of stress in parent-child relationships. The measure should be completed by the primary caregiver about the child in your family who has 18q-. The PSI is comprised of 120 items and typically takes about 20 minutes to complete. Please use the PSI Answer Sheet to mark your responses to questions from the PSI Item Booklet. Please be sure to use a pen for this form.

The FES (Form R) should also be completed by a primary caregiver about his/her actual perceptions of the family environment. The FES is comprised of 90 true/false items and typically takes about 20 minutes to complete. Please mark your answers to the Item Booklet questions in the grid on the back of the FES Answer Sheet. Please also note that the items in the Item Booklet are written across the page, so be sure to match the number of the question in the booklet with the number of your response on the answer sheet.

We would also like for you to complete the demographic information on both forms, including your name and your child's birth date. When complete, please use the enclosed SASE to return to Bridgette Soileau.

Thank you for your willingness to participate in our mail out survey.

Sincerely,

Jannine D. Cody, Ph.D.	Daniel E. Hale, MD
Principal Investigator	Medical Director
The Chromosome 18 Clinical Research Center	The Chromosome 18 Clinical Research Center

Appendix D: Information Letter
Control Participants

COLLEGE OF EDUCATION
THE UNIVERSITY OF TEXAS AT AUSTIN

Department of Educational Psychology • George I. Sánchez Building 504 • Austin, Texas 78712-1296
(512) 471-4155 • FAX (512) 471-1288 • Campus Mail Code D5800 • http://www.edb.utexas.edu/coe/depts/edp

Dear Families,

My name is Kim Davis, and I'm a 5th year doctoral student in the School Psychology program at the University of Texas at Austin. I am compiling data for my dissertation. In conjunction with the Chromosome 18 Clinical Research Center at the UT Health Science Center, my study will examine the influences of genetic disorders such as 18q- and Down Syndrome on various aspects of family functioning. Thank you for volunteering to participate in this developmental mail out survey on the topic of parenting stress and family environment.

Enclosed you will find a series of consent forms that you will need to sign in order to participate in this study. The consent forms detail our privacy and confidentiality policies, as well as a description of the minimal risks associated with participating. Please be sure to include the consent forms when you return the packet.

You will also find a copy of the Parenting Stress Index (PSI) and the Family Environment Scale (FES). The PSI is a self-report measure that provides an estimate of areas of stress in parent-child relationships. The measure should be completed by the primary caregiver about one child in your family who is between the ages of 1 month and 6 years. The PSI is comprised of 120 items and typically takes about 30 minutes to complete. Please use the PSI Answer Sheet to mark your responses to questions from the PSI Item Booklet. Please be sure to use a pen for this form.

The FES (Form R) should also be completed by a primary caregiver about his/her actual perceptions of the family environment. The FES is comprised of 90 true/false items and typically takes about 20 minutes to complete. Please mark your answers to the Item Booklet questions in the grid on the back of the FES Answer Sheet. Please note that the items in the Item Booklet are written across the page, so be sure to match the number of the question in the booklet with the number of your response on the answer sheet.

We would also like for you to complete the demographic information on both forms, including your name, ethnicity, marital status, and the birthday of your child who is between the ages of 1 month and 6 years. When complete, please use the enclosed self-addressed, stamped envelope to return the forms.

Thank you very much for your willingness to participate in this mail out survey.

Sincerely,

Kim S. Davis, M. A.
Doctoral Student
University of Texas at Austin

Margaret Semrud-Clikeman, Ph.D.
Professor
University of Texas at Austin

Appendix E: Information Letter
Down Syndrome Participants

COLLEGE OF EDUCATION
THE UNIVERSITY OF TEXAS AT AUSTIN

Department of Educational Psychology • George I. Sánchez Building 504 • Austin, Texas 78712-1296
(512) 471-4155 • FAX (512) 471-1288 • Campus Mail Code D5800 • http://www.edb.utexas.edu/coe/depts/edp

Dear Families,

My name is Kim Davis, and I'm a 5th year doctoral student in the School Psychology program at the University of Texas at Austin. I am compiling data for my dissertation. In conjunction with the Chromosome 18 Clinical Research Center at the UT Health Science Center, my study will examine the influences of genetic disorders such as 18q- and Down Syndrome on various aspects of family functioning. Based on your child's age and diagnosis of Down Syndrome, you have been selected to participate in a developmental mail out survey on the topic of parenting stress and family environment.

Enclosed you will find a series of consent forms that you will need to sign in order to participate in this study. The consent forms detail our privacy and confidentiality policies, as well as a description of the minimal risks associated with participating. Please be sure to include the consent forms when you return the packet.

You will also find a copy of the Parenting Stress Index (PSI) and the Family Environment Scale (FES). The PSI is a self-report measure that provides an estimate of areas of stress in parent-child relationships. The measure should be completed by the primary caregiver about the child in your family who has Down Syndrome and is between the ages of 1 month and 6 years. The PSI is comprised of 120 items and typically takes about 30 minutes to complete. Please use the PSI Answer Sheet to mark your responses to questions from the PSI Item Booklet. Please be sure to use a pen for this form.

The FES (Form R) should also be completed by a primary caregiver about his/her actual perceptions of the family environment. The FES is comprised of 90 true/false items and typically takes about 20 minutes to complete. Please mark your answers to the Item Booklet questions in the grid on the back of the FES Answer Sheet. Please note that the items in the Item Booklet are written across the page, so be sure to match the number of the question in the booklet with the number of your response on the answer sheet.

We would also like for you to complete the demographic information on both forms, including your name, ethnicity, marital status, and the birthday of your child who has Down Syndrome. When complete, please use the enclosed self-addressed, stamped envelope to return the forms.

Thank you very much for your willingness to participate in this mail out survey.

Sincerely,

Kim S. Davis, M. A.
Doctoral Student
University of Texas at Austin

Margaret Semrud-Clikeman, Ph.D.
Professor
University of Texas at Austin

Appendix F: IRB Consent Form

IRB # 2001-04-0015

Informed Consent to Participate in Research

The University of Texas at Austin

You are being asked to participate in a research study. This form provides you with information about the study. The Principal Investigator (the person in charge of this research) or his/her representative will also describe this study to you and answer all your questions. Please read the information below and ask questions about anything you don't understand before deciding whether or not to take part. Your participation is entirely voluntary and you can refuse to participate without penalty or loss of benefits to which you are otherwise entitled.

Title of Research Study: The Chromosome 18 Clinical Research Center

Principal Investigator(s): Professor Margaret Semrud-Clikeman, Ph.D., Department of Educational Psychology, School Psychology Program, University of Texas at Austin, (512) 471-0274

Funding Source: None

What is the purpose of this study?
This study is an amendment to a larger study about the implications of Chromosome 18 disorders. This portion of the study focuses on families of children with Down Syndrome and children without any clinical diagnoses. We are trying to learn about the family environment and parenting stress in families of young children with genetic disorders, as well as in families with typically developing young children. The extensive impairments associated with genetic disorders, both physical and behavioral, may be a significant source of stress to parents. We would like to determine what types of environments exist within these families, as well as what types of stress and the amount of such stress that parents experience. We are asking parents of typically developing children to participate in this study in order to determine what differences take place when parenting a child with a genetic disorder. You will be one of approximately 90 parents asked to participate in the project over the next year.

What will be done if you take part in this research study?
You will be asked to complete two self-report forms that take approximately 45 minutes to complete all together. On these forms, you will be asked to answer a set of demographic questions, including your ethnicity, marital status, and child's birthday. You will also be asked several multiple choice and true/false questions about stressors you may experience related to parenting and family relationships.

What are the possible discomforts and risks?
There are few known risks to this study. You may feel uncomfortable with the personal nature of some of the questions asked on the self-report forms.

What are the possible benefits to you or to others?
There are no significant benefits of participating in the study for you or your child.

If you choose to participate in this study, will it cost you anything? No

Will you receive compensation for your participation in this study? No

What if you are injured because of the study?
There are no known physical risks. No treatment will be provided for research related injury and no payment can be provided in the event of a medical problem.

If you do not want to take part in this study, what other options are available to you?
Participation in this study is entirely voluntary. You are free to refuse to be in the study, and your refusal will not influence your current or future relationship with The University of Texas at Austin.

How can you withdraw from this research study and who should you call if you have questions?
If you wish to stop your participation in this research study for any reason, you should contact Margaret Semrud-Clikeman, Ph.D. (512) 471-0274. You are free to withdraw your consent and stop participation in this research study at any time without penalty or loss of benefits for which you may be entitled. Throughout the study, the researchers will notify you of new information that may become available and that might affect your decision to remain in the study.

In addition, if you have questions about your rights as a research participant, **please contact Lisa Leidin, Ph.D., Chair, The University of Texas at Austin Institutional Review Board for the Protection of Human Subjects, (512) 471-8871.**

How will your privacy and the confidentiality of your research records be protected?
Authorized persons from The University of Texas at Austin and the Institutional Review Board have the legal right to review your research records and will protect the confidentiality of those records to the extent permitted by law. If the research project is sponsored then the sponsor also has the legal right to review your research records. Otherwise, your research records will not be released without your consent unless required by law or a court order.

If the results of this research are published or presented at scientific meetings, your identity will not be disclosed.

The forms completed by participants will be kept in a secure place (e.g., a locked file cabinet in the investigator's office) and will be viewed only for research purposes by the investigator and his or her associates.

Will the researchers benefit from your participation in this study? No

Signatures:

As a representative of this study, I have explained the purpose, the procedures, the benefits, and the risks that are involved in this research study:

_____

Signature and printed name of person obtaining consent **Date**

You have been informed about this study's purpose, procedures, possible benefits and risks, and you have been given the option to make a copy of this form. You have been given the opportunity to ask questions before you sign, and you have been told that you can ask other questions at any time. You voluntarily agree to participate in this study. By signing this form, you are not waiving any of your legal rights.

_____

Printed Name of Participant **Date**

_____

Signature of Participant **Date**

_____

Signature of Principal Investigator **Date**

CONSENT FORM

The Chromosome 18 Clinical Research Center

You are invited to participate in a study of several variables of parenting stress and family environment in families of young children with and without genetic disorders. My name is Margaret Semrud-Clikeman, Ph.D., and I am a professor at The University of Texas at Austin, Department of Educational Psychology. I am asking for permission to include you and your child in this study because we are examining family environment and parenting stress in families of children with and without genetic disorders. We are working with families who have young children with genetic disorders, as well as with families who do not. I expect to have 90 participants in this study.

If you agree to participate, Margaret Semrud-Clikeman, Ph.D. will discuss the types of forms you will complete. These forms include completing demographic information and answering questions about your family and your beliefs. Completion of these forms will take place in your own home at your convenience.

Any information that is obtained in connection with this study and that can be identified with you or your child's name will remain confidential and will be disclosed only with your permission. Your responses will not be linked with your or your child's names in any written or verbal report of this research project. No information will be released without written permission from you.

Your decision to participate will not affect you or your family's present or future relationship with The University of Texas at Austin. If you have any questions, please ask me. If you have any questions later, please call me at (512) 471-0274. If you have any questions about your participation in this study, call Professor Clarke Burnham, Chair of The University of Texas at Austin Institutional Review Board for the Protection of Human Research Participants at (512) 232-4383.

You may keep a copy of this consent form.

You are making a decision about participating in this study. Your signature below indicates that you have read the information provided above and have decided to participate in the study. If you later decide that you wish to withdraw your permission for your participation in the study, simply tell me. You may discontinue your participation at any time.

_____
Printed Name of Primary Caregiver

_____ _____
Signature of Primary Caregiver Date

_____ _____
Signature of Investigator Date

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VITA

Kim Suzanne Davis was born in Houston, Texas on February 10, 1979, the daughter of Robert Davis and Arlene Friedman Davis. After receiving her diploma at The Kinkaid School, Houston, Texas, 1997, she entered Colby College in Waterville, Maine. In January of 1999, she transferred to the University of Texas at Austin, where she received the degree of Bachelor of Arts in Psychology in May, 2001. In August, 2001, she entered the doctoral program in School Psychology at the University of Texas at Austin. She received a Master of Arts from the University of Texas in August, 2004. She will complete her pre-doctoral internship at Columbus Children's Hospital and the Ohio State University in August 2007. In September 2007, she will begin a post-doctoral year at Children's Medical Center of Dallas.

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This dissertation was typed by the author.